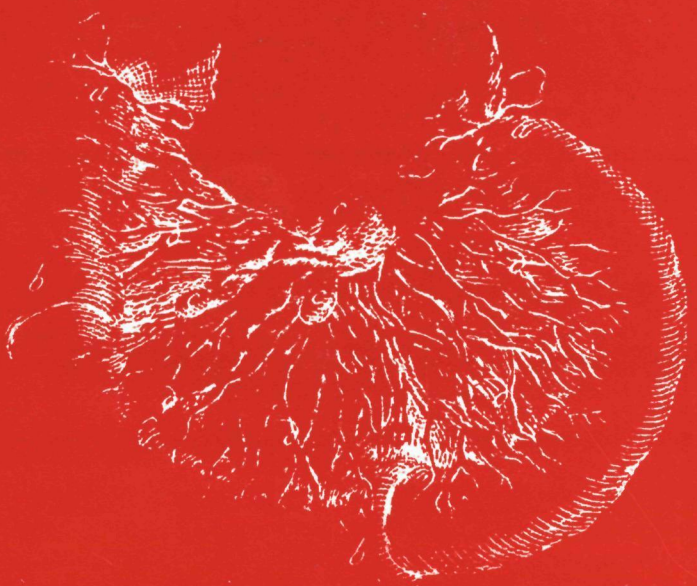
INTUSSUSCEPTION

a clinical and experimental study



J.A.M. REIJNEN

INTUSSUSCEPTION

a clinical and experimental study

Promotor: Prof. Dr. C. Festen

Co-promotor: Dr. H.J.M. Joosten

INTUSSUSCEPTION

a clinical and experimental study

een wetenschappelijke proeve op het gebied
van de geneeskunde en de tandheelkunde

Proefschrift

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Aggressiveness helps social animals to provide and maintain a hierarchic order so that the strongest and most talented animals have to devote themselves hardest for the community. N.N.

To my parents

Contents

Chapter I	**INTRODUCTION - AIM OF THE STUDY**	1
Chapter II	**GENERAL PART - REVIEW OF LITERATURE**	3
II.1.	HISTORY	3
II.2.	NOMENCLATURE	9
II.3.	CLASSIFICATION	9
II.3.1.	**Classification according to direction**	9
II.3.2.	**Classification according to development**	11
II.3.3.	**Classification according to localization**	11
II.3.4.	**Classification according to duration of symptoms**	13
II.4	EPIDEMIOLOGY	14
II.4.1.	**Incidence**	14
II.4.2.	**Geographical variation**	15
II.4.3.	**Seasonal incidence**	15
II.4.4.	**Distribution according to gender**	16
II.4.5.	**Racial incidence**	17
II.4.6.	**Heredity**	17
II.4.7.	**Distribution according to age**	17
II.4.7.1.	Intra-uterine intussusception	18
II.4.7.2.	Intussusception in neonates	19
II.5.	INTUSSUSCEPTION IN OTHER SPECIES	20
II.6.	ETIOLOGY OF INTUSSUSCEPTION	21
II.6.1.	**Factors related to the contraction state of the bowel**	22
II.6.1.1.	Paralysis	22
II.6.1.2.	Spasm	22
II.6.1.3.	Autonomic nervous system dysfunction	23
II.6.2.	**Factors related to local anatomic characteristics**	24
II.6.2.1.	Appendiceal intussusception	25
II.6.3.	**Local mechanical factors**	27

II.6.3.1.	Meckel's diverticulum	27
II.6.3.2.	Cysts	28
II.6.3.3.	Heterotopic tissue	28
II.6.3.4.	Benign tumours	29
II.6.3.5.	Malignant tumours	29
II.6.3.6.	Henoch-Schönlein purpura	30
II.6.3.7.	Mucoviscidosis	31
II.6.3.8.	Bezoars	31
II.6.4.	**Factors related to infection and inflammation**	32
II.6.4.1.	Viral infection	32
II.6.4.2.	Lymphoid hyperplasia	34
II.6.4.3.	Bacterial and parasitic infection	36
II.6.4.4.	Inflammatory bowel disease	37
II.6.5.	**Miscellaneous factors**	37
II.6.5.1.	Intestinal intubation	37
II.6.5.2.	Trauma	38
II.6.5.3.	Postoperative intussusception	39
II.6.5.4.	Intussusception after jejuno-ileal bypass	41
II.6.5.5.	Intussusception after gastro-jejunostomy	42
II.6.5.6.	Intussusception in chronic renal failure	43
II.6.5.3.	Intussusception in pregnancy	44
II.7.	SYMPTOMATOLOGY	44
II.7.1.	**Cardinal symptoms and signs**	45
II.7.1.1.	Abdominal pain	45
II.7.1.2.	Vomiting and anorexia	46
II.7.1.3.	Rectal blood loss	47
II.7.1.4.	Tumour and prolapse	48
II.7.2.	**Accessory symptoms and signs**	49
II.7.2.1.	General condition	49
II.7.2.2.	Constipation and diarrhoea	50
II.7.2.3.	Anorexia and weight loss	51
II.7.2.4.	Upper respiratory tract infection and otitis media	51
II.7.2.5.	Signs of bowel obstruction	51
II.7.2.6.	Miscellaneous symptoms and signs	52
II.8.	DIAGNOSTICS	52
II.8.1.	**Laboratory tests**	52
II.8.2.	**Imaging studies**	53
II.8.2.1.	Plain abdominal radiograph	53
II.8.2.2.	Contrast enema examination	54
II.8.2.3.	Small bowel follow-through meal	56
II.8.2.4.	Computed tomography	57
II.8.2.5.	Other imaging techniques	57

II.9. DIFFERENTIAL DIAGNOSIS 58

II.10. TREATMENT OF INTUSSUSCEPTION 60
II.10.1. **Operative treatment** 60
II.10.2. **Conservative treatment** 63
II.10.2.1. Spontaneous reduction 63
II.10.2.2. Sloughing of the intussusceptum 63
II.10.2.3. Hydrostatic reduction 64
II.10.2.4. Pharmacological support of reduction 70
II.10.2.5. Gasreduction of intussusception 72

II.11. COMPLICATIONS 73
II.11.1. **Mortality** 73
II.11.2. **Bowel perforation** 74
II.11.3. **General complications** 74
II.11.4. **Recurrent intussusception** 75

II.13. REFERENCES 78

Chapter III **OWN OBSERVATIONS AND INVESTIGATIONS** 101

III.1. ATYPICAL CHARACTERISTICS OF A GROUP OF CHILDREN WITH INTUSSUSCEPTION.
ACTA PAEDIATRICA SCANDINAVICA, accepted for publication. 103

III.2. INTUSSUSCEPTION: FACTORS RELATED TO THE OUTCOME OF TREATMENT.
ARCHIVES OF DISEASE IN CHILDHOOD, accepted for publication. 111

III.3. NEUROLOGIC SIGNS AS EARLY MANIFESTATIONS OF INTUSSUSCEPTION IN CHILDREN.
AMERICAN JOURNAL OF DISEASES OF CHILDREN, accepted for publication. 125

III.4. CHRONIC INTUSSUSCEPTION IN CHILDREN.
BRITISH JOURNAL OF SURGERY, 1989; 76: 815-816. 135

III.5. INTUSSUSCEPTION IN CHILDREN 5-15 YEARS OF AGE.
BRITISH JOURNAL OF SURGERY, 1987; 74: 692-693. 141

III.6. DIAGNOSIS AND TREATMENT OF ADULT INTUSSUSCEPTION.
AMERICAN JOURNAL OF SURGERY, 1989; 158: 25-28. 149

III.7. INTUSSUSCEPTION COMPLICATED BY BOWEL PERFORATION DURING HYDROSTATIC REDUCTION.
ZEITSCHRIFT FÜR KINDERCHIRURGIE, accepted for publication. 159

III.8. SONOGRAPHIC FEATURES OF INTUSSUSCEPTION.
RADIOLOGY, submitted. 167

III.9. INTRAPERITONEAL INJECTION OF BORDETELLA PERTUSSIS VACCINE AND THIOMERSAL FAILED TO PRODUCE INTUSSUSCEPTION IN MICE.
ZEITSCHRIFT FÜR VERSUCHSTIERKUNDE, 1988; 31: 245-247. 181

Chapter IV **SUMMARY AND CONCLUSIONS** 187

ACKNOWLEDGEMENTS 203

CURRICULUM VITAE 205

CHAPTER I

Introduction – Aim of the study

Intussusception of the bowel has interested investigators since the time of Hippocrates. Nevertheless many obscurities still exist. Still no answer can be given to the question why the incidence of intussusception is highest during the first two years of life. Neither is there any explanation for the male preponderance that is reported in all series of intussusception in children. Moreover we do not know with certainty why most intussusceptions occur in the ileocecal region. Small transient intussusceptions have been occasionally noticed during laparotomy. This phenomenon suggests the prevalence of a "physiologic" intussusception but its incidence and clinical appearance have never been established. Although many theories as to the pathogenesis of intussusception have been proposed, the most intriguing problem remains the high incidence of intussusception in children without an obvious cause. One of the reasons why there are so many questions still to be answered, may be the fact that we do not have a proper animal model to study intussusception of the bowel.

Great progress has been made in the treatment of intussusception in children. At the end of the last century an 80% mortality from operation was usual. Nowadays no child succumbs to the effects of an intussusception unless it is admitted in an already moribund state. Primary operative treatment is replaced by a conservative approach with an attempt at hydrostatic reduction. Controversy still exists concerning the question if all cases of intussusception should be treated primarily hydrostatically. There is no clarity under which circumstances unnecessary delay of resection of nonviable or pathologic bowel and the risk of bowel perforation by hydrostatic reduction attempt can be avoided through primary surgical therapy. The correct way of hydrostatic reduction is another point of debate. There is no unanimity concerning manipulation of the intussusception through the abdominal wall and concerning the height to which the barium canister can be raised. Also the number of attempts and the time to be spent for these attempts, is a matter in dispute. The medication, used to improve the success rate of hydrostatic reduction, has been the subject of too few studies. Recently the advantages of air instead of barium for reduction have been outlined.

In the last two decades computed tomography, nuclear scanning, endoscopy and ultrasonography have been described as diagnostic tools for intussusception. For routine use, only ultrasonography is a readily available, valuable instrument in experienced hands. However, early diagnosis, associated with a favorable outcome of treatment, still depends on the recognition of the clinical features of intussusception. More and more an important number of intussusceptions is reported to show other characteristic symptoms and signs than those described in the past.
The incidence of intussusception in the Netherlands is about 250 patients a year. The experience with this uncommon but not rare clinical entity of many general practitioners but also of many consulting physicians must be limited. Delay in diagnosis and treatment, due to inexperience, may lead to (unnecessary) mortality and morbidity. One of the aims of this study was to draw attention to this life threatening but, in an early stage, easily curable disease. The study intends to provide a review of the literature data concerning intussusception in children as well as adults and to present results of own investigations in patients with intussusception and in animals, with special emphasis on the etiological, clinical, diagnostic and therapeutical aspects of this disorder.
The questions this study aims to answer are:

1. Which clinical features of intussusception bare diagnostic significance? Which clinical features of intussusception are important from a diagnostic point of view for different age groups?

2. Can we provide guidelines on the choice of treatment of intussusception on the ground of clinical features? Must we provide different guidelines on the choice of treatment of intussusception for different age groups?

3. What can be the contribution of ultrasonography to the diagnosis of intussusception? What can be the contribution of ultrasonography to guidelines on the choice of treatment of intussusception?

4. Can we develop a practical, cheap and "physiologic" animal model to study intussusception?

CHAPTER II

General part – Review of literature

II.1. HISTORY

In the times of Hippocrates and Praxagoras intussusception was included in the term "ileos" (ileus). This term covered diseases in the organs of the abdominal cavity of the most diverse kinds. Yet, all they had in common was that they were considered to have originated in inflammatory processes. Thus, the mechanical type of ileus was unknown. Hippocrates advised forced injection of water or air into the intestine in all forms of ileus. (The air was administered by means of bellows without any control of the pressure). In contradistinction Praxagoras is said to have advocated opening the abdominal cavity to restore the normal situation (164).

About the end of the sixteenth century and the beginning of the seventeenth Realdus Columbus, Fabricius Hildanus and Riolan produced an anatomical description of invagination. In 1627 the French surgeon Francois Rauchin suggested that intussusception should be excluded from the collective term ileus. In 1674 Paulus Barbette of Amsterdam described in his "Chirurgie"

> *Het Gedarmte wringht hem gelijck een worm, en dan geschiet het somtijdts (insonderheydt in heftige pijnen) dat het in malkander schiet, en alsoo de vuyligheden haren doorgang beneemt. Dese siekten heet men Ileos, of Miserere mei. Als zy op de gewoone middelen niet wycken wil, soude een groote loose kop dickmaels opgeset en afgetrocken, wat voordeel konnen doen: was het oock niet beter dat men een opening door de Spieren en de Pens-sack maeckte, en met de vingeren het gedarmte uyt malkanderen trock; als dat men de siecke liet verlooren gaen?*
>
> The Bowel wriggles itself like a worm, and then it comes to pass that sometimes (in particular in fierce pains) it shoots into itself, and thus hindering the dirts its passage. This disease is named Ileos, or Miserere mei. If it does not yield to the usual remedies, a large cupping-glass, frequently put on and withdrawn, may be of benefit: would it not be better to cut open the muscles and the

peritoneum and pull the bowel out of itself with one's fingers than to lose the diseased person? (12).

This is the first recorded suggestion of operative reduction.
The first case of a successful operation on an intussusception was described by Velse of Leyden in his thesis "De mutuo intestinorum ingressu et aliis machinae humanae extraordinariis" in 1742.

Quum nimirum foemina quinquagenaria, admodum truculentis hujus morbi symptomatis defessa, a nulla remedio Enematum, Fomentorum, Cataplasmatum, & cucurbitarum magnarum ventosarum abdominis, a celeberrimo Nukkio successive applicatorum, levamen ullum cepisset; Ille, in praxi felicissimus, suspicatus intestinorum mutuum amplexum subesse, author fuit, ut Chirurgus dexterrimus, in latere sinistro, quator digitos transversos ab umbilico, oblique versus inferiora simul & posteriora facto abdomini hiatu, intestina, (statim absque ulla mora lacte tepido fovenda) protraheret, quaereret locum involutum, eumque lente evolveret, mox omnia intestina reponeret, & tandem vulnus abdominis consueret. Quod consilium dictum factum adeo bene cessit huic, fere jam morienti, mulieri, ut eductis intestinis, &, bona fortuna, subito eorum immersione, omnium malorum matre, reperta, inflamata necdum, nec coalita, & post olei multi inunctionem diducta; atque postremo, post eorum refusionem secundum artem, gastrorapbia ex voto absoluta; primum quidem Enemata Emollienta alvum denuo aperuerint, postea vero vires restaurati corporis ipsae sponte sua apertam illam servaverint, eo effectu, ut Illa ex Orci faucibus veluti erepta brevi perfecte sanata, & ultra 20. deinceps annos superstes, fuerit.

A woman of quite fifty years, exhausted by the fell symptoms of this disease, had received no benefit from any remedy for the great distension of the abdomen, whether enemata, fomentations or cataplasms which had been tried by the celebrated Nuck; whereupon the latter, a very skilled practitioner, suspecting that there was an intussusception, advised a very skilful surgeon to make an incision in the left side of the abdomen, extending about four fingers breadth downward and backward from the umbilicus. The surgeon was then to draw out the intestines immediately bathing them in tepid milk, seek for the affected part and gently reduce it. Thereupon he should replace the intestines and finally sew up the wound. This advise was so well carried out that it happened in the case of this already moribund woman that the intestines were brought out and by good fortune, on their immersion the source of the trouble was found immediately; there were

no adhesions nor any inflammation, and after skilful suture the abdominal wound healed satisfactorily. At first emollient enemata opened the bowels, but after that the renewed bodily strength spontaneously performed that act so satisfactorily that, snatched as it were from the jaws of death, she soon recovered perfect health and lived thereafter more than twenty years. (353)

This case was an exception as Dutch physicians in the eighteenth century used to apply a wide variety of conservative measurements. Besides purging, enema's, blood-letting, oral administration of quicksilver or leaden bullets the tobacco smoke clyster was advocated (223). In 1741 in his translation of Heister's famous book 'Chirurgie', Ulhoorn described the successful application of tobacco smoke.

Ten tijde als myn Vaderlyke vriend, den uitmuntende Professor F. Ruysch nog leefde, zag ik met hem een diergelyke lyder als zieltogende, hebbende reets verscheide dubbelde en drievoudige purgatien beneffens drie koegels in het lichaam, zonder dat 'er eenige ontlasting volgde, om welke reden wy in de tyd van een uur twee tabaks-klisteren zetteden, die van zulk een goede uitwerking waren, dat wanneer den lyder op het stilletje zat, ons niet anders deed geloven of hy zoude met zyne drie koegels die hy loosde, den pot hebben in stukken geschoten, en dus wierd deze miserie in een vrolyk gelaat verandert.

At the time when my paternal friend, the excellent Professor F. Ruysch was still alive, I saw with him such a patient who seemed to be a dying man, having had already several double and triple purgations together with three bullets in his body without any motions following, for which reason we applied two tobacco-enemas within the space of one hour which was so effective that when the patient was sitting on the night-stool, he made us all but believe that by the forcible discharge of his three bullets, he would have shot the chamber-pot to pieces and thus this misery turned into a merry countenance (351).

Rigail of Leyden was the first to use the term intussusception. In his thesis of 1769 'De intestinorum intussusceptu', he stated that the beginning of an intussusception was a spastic contraction of the bowel caused by unsound food (287). A statement that still can well be maintained nowadays. These facts speak well for the high standard of medical science in the Netherlands in the 17th and 18th century. The fine drawing of a double intussusception in Eduard

Sandiforts 'Observationes anatomico-pathologicae' is another witness (see front page illustration) (300).

The conservative form of treatment also remained the first choice in the 19th century. Patients were transferred into the hands of the surgeon only when they were in a fatal condition. This fact contributed seriously to the high mortality figures of surgical intervention.

In 1874 sir Jonathan Hutchinson reported the first successful operation on a child. Conservative means had failed to reduce a chronic ileocoecocolic intussusception that protruded from the anus. Hutchinson reduced it digitally during an operation that did not occupy more than two or three minutes. The child made an uneventful recovery (155).

In 1876 Hirschsprung of Kopenhagen published the first of a series of reports dealing with the systematic reduction of intussusception by hydrostatic reduction (141). His results were far superior to those achieved by primary operative treatment. At first he was not believed but after his report of 107 cases in 1905, his technique was adopted in many countries (142).
With the introduction of antiseptics at the end of the eighteen-eighties, abdominal surgery entered a new era. The mortality fell enormously while, simultaneously, the surgical experience rose inversely. In 1897 Clubbe did the first successful resection of an intussusception in a child (46). Since that time the treatment of intussusception was subject of much debate again.

In 1913 Ladd published the first case of a colonic intussusception diagnosed by means of contrast enema and in 1918 Langley Porter suggested the use of the contrast enema for the purpose of controlled reduction (46, 239). Over the next half century little by little this therapeutical mode gained acceptance as the treatment of first choice. Recently in the western literature several reports from the Far East described the successful reduction of intussusception by pressure controlled air insufflation (124, 339, 377, 385). History repeats itself.

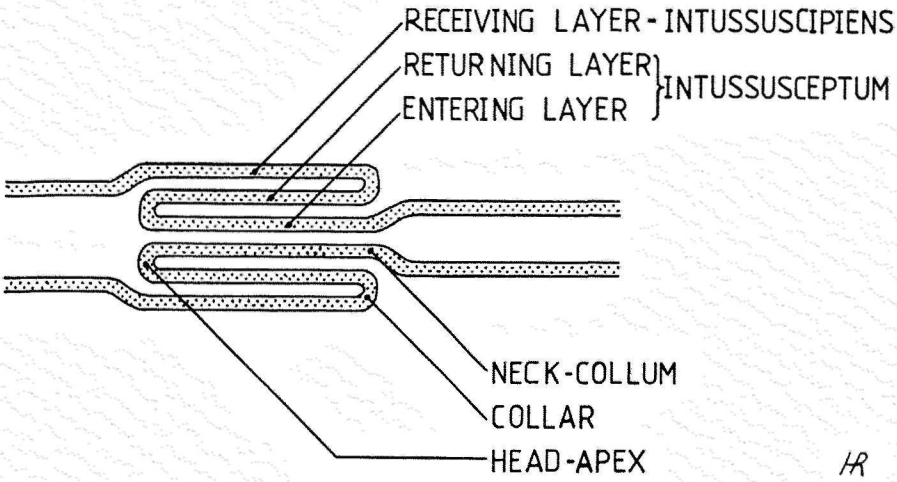

Fig. 1. Anatomical features of intussusception.

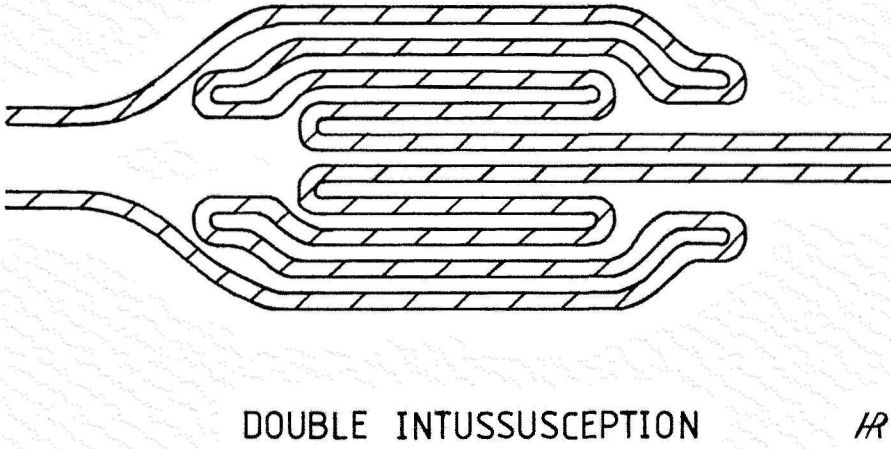

Fig. 2. Diagram of double, complex or telescopic intussusception.

The term intussusception applies to the introversion of a portion of the intestine into another, adjacent part. An intussusception consists of three concentric cylinders, each cylinder consisting of the full thickness of the intestinal wall (Figure 1). The outermost cylinder is called intussuscipiens or invaginans (English: the sheath or the receiving layer; French: la gaine; German: der Scheide; Dutch: de schede). The two inner cylinders together form the intussusceptum or invaginatum (French: le boudin). In the English literature the middle cylinder is called the returning layer and the innermost cylinder is called the entering layer. The foremost part of the intussusceptum, i.e. the precedent one, is termed apex or caput (English: the head; French: la tête (fixe); German: der Spitze), it is the transition from the entering layer to the returning layer. The turn-back of the returning layer to the receiving layer is called collar (French: le bourrelet ou le collet (mobile); German: der Kragen; Dutch: de kraag). The part of the entering layer enfolded by the collar is the collum (English: the neck; Dutch: de hals).
When an intussusception is formed by three intestinal wall layers, it is called simple. Also double or complex, so-called telescopic, forms occur (Figure 2). Thus, a double intussusception consists of five cylinders, a treble one of seven. As a rule intussusceptions are single although cases with multiple intussusceptions are not rare.

According to their mode of origination, intussusceptions are divided into agonal and vital ones. The agonal intussusceptions will be discovered, not infrequently, in connection with postmortems of both children and adults. They are easily differentiated from those arising intra vitam, through their insignificant extent and the absence of circulatory disturbances. This relates them to so called 'physiologic' intussusceptions assumed to happen in every person every day without giving any complaints or problems. They might well be similar with the small transient intussusceptions seen during laparotomy for other reasons.

II.3. CLASSIFICATION

II.3.1. Classification according to direction

Usually, the course of an intussusception is a descending one, i.e. antegrade or isoperistaltic, in so far as an orally placed intestinal portion will be invaginated in one situated in an anal (aboral) direction next to it. Ascending forms are called retrograde. Most

retrograde intussusceptions occur after intestinal intubation or gastroenterostomy. Spontaneous retrograde intussusceptions are very rare. Most intussusceptions are central or total, i.e. in the direction of the axis of the intestine. Lateral or partial intussusceptions are (rarely) described in the cecum.

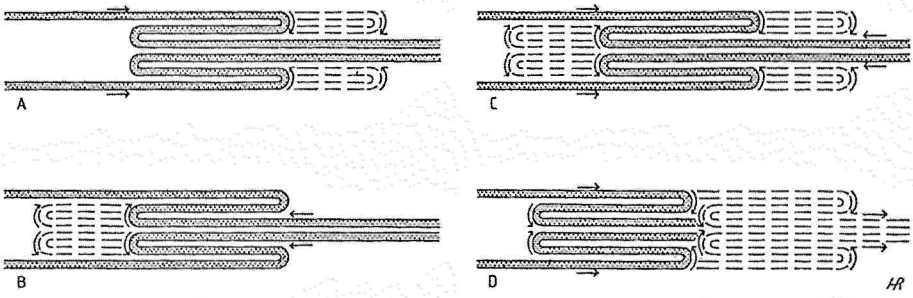

Fig. 3a,b,c and d. Classification of intussusception according to development.

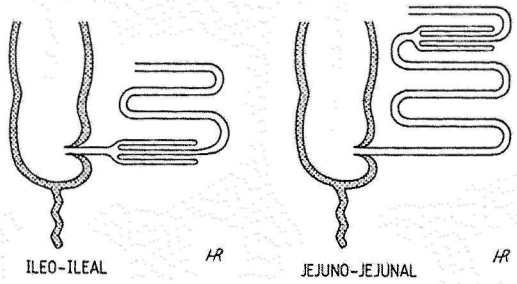

Fig. 4 and 5. Jejuno-jejunal and ileo-ileal intussusception.

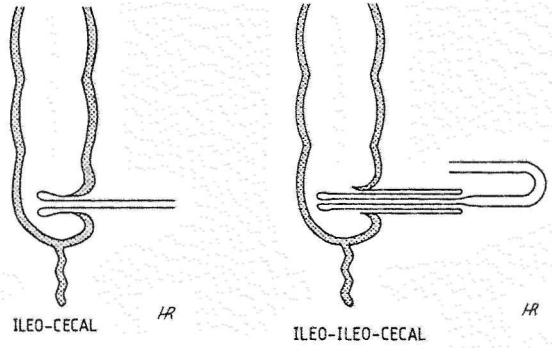

Fig. 6. Ileo-cecal and ileoileo-cecal intussusception. retrograde or antiperistaltic.

II.3.2. Classification according to development

Intussusceptions are also classified according to the way they develop. Four types are distinguished. In the first type the apex of the intussusception is always constant, i.e. it will at all stages be identical to the intestinal portion that was first introverted. These intussusceptions increase at the expense of its sheath. The collar is formed by different parts of the intussuscipiens at different stages (Figure 3a). In the second type the collar of the intussusception remains constant i.e. it will at all stages be identical to the intestinal portion that first received the intussusceptum. These intussusceptions increase at the expense of the proximal intestine. The apex is formed by different parts of the entering layer (Figure 3b). The third type of intussusception increases at the cost of both receiving layer and proximal intestine and so is a combination of the first two types (Figure 3c). The fourth type remains constant by increasing at the expense of its sheath and by at the same time decreasing at the apex in favour of the entering layer, in this way changing its localization along the intestinal tract (Figure 3d).

II.3.3. Classification according to localization

Intussusceptions of all parts of the digestive tract have been reported but also of the biliary system, the ureter and the Fallopian tube (2). Evidently it takes a movable, hollow tube with a smooth muscle layer to produce an intussusception. In theory an intussusception of the ductus deferens is possible but to my knowledge never reported. An intussusception is named after the particular intestinal portion that has first appeared as intussusceptum and according to its position within the intestinal canal. Different authors classify the same forms of intussusception under different headings or apply different names to the same type of intussusception. The following is a clear, easy to understand and well accepted classification of the most frequent intussusceptions (370).

A. Small intestinal or enteric intussusceptions

1. Jejuno-jejunal intussusceptions (Figure 4)
2. Ileo-ileal intussusceptions (Figure 5)

B. Intussusceptions at the boundary between the small intestine and the colon.

1. Ileo-(ileo-)cecal intussusceptions (Figure 6)
2. Ileo-(ileo-)colic intussusceptions (Figure 7)

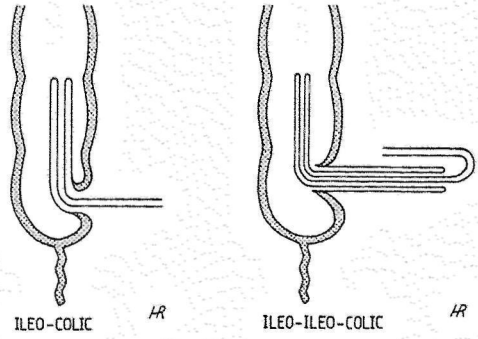

Fig. 7. Ileo-colic and ileoileo-colic intussusception.

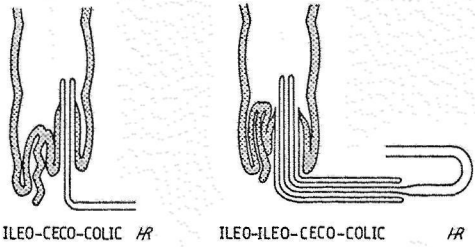

Fig. 8. Ileoceco-colic and ileoileo-ceco-colic intussusception.

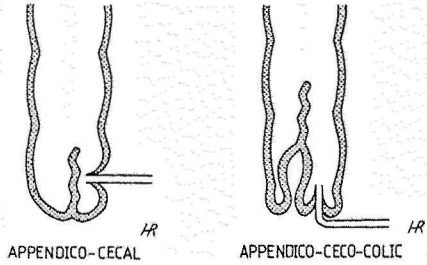

Fig. 9. Appendico-cecal and appendico-ceco-colic intussusception.

3. Ileo-(ileo-)ceco-colic intussusceptions (Figure 8)
4. Appendico-cecal/appendico-ceco-colic intussusceptions (Figure 9)
5. Ceco-colic intussusceptions (Figure 10)

C. Colic intussusceptions.

1. Colo-colic intussusceptions (Figure 11)

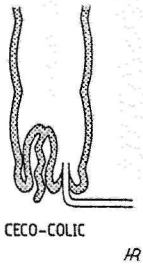

Fig. 10. Ceco-colic intussusceptions

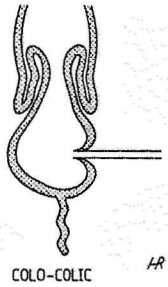

Fig. 11. Colo-colic intussusceptions

Mixed intussusceptions by combination of different types, ante- and retrograde intussusceptions and by doubling have been described.
The distribution of intussusceptions according to localization is different in different age groups. In series concerning children under 15 years of age with a common age distribution, that is to say about 75% of children under the age of 2 years, between 5 and 10% of intussusceptions are enteric, between 85 and 90% of intussusceptions are at or around the ileocecal valve and the rest, usually less than 5%, is of the colocolic type (9, 11, 62, 129, 174, 187, 203, 239, 249, 256, 273, 315, 337). In series concerning both children and adults (164, 240, 259) the percentage of enteric intussusceptions is between 10 and 20. Also the percentage of colic intussusceptions is higher, 8 to 13. In children under the age of 1 year intussusceptions of the enteric type are less frequent (<5%) and so are colic intussusceptions (164, 239, 370). Though intussusceptions at the boundary between the small and the large intestine predominate in all age groups the enteric and colic intussusceptions are more common among older children and adults (164, 228, 350, 370).

II.3.4. Classification according to duration of symptoms

The classification with a view to the clinical course dates back from 1878 (268). Intussusceptions causing death within 1-2 days are

called peracute. Intussusceptions leading to death within 3-7 days are acute. Subacute intussusceptions are lethal within 8-14 days. Chronic intussusceptions last for more than two weeks. It must be emphasized that this classification is unfit for present conditions, as intussusceptions nowadays are diagnosed and treated earlier than before. A classification into acute and chronic intussusceptions will suffice. An intussusception of an acute course produces a complete obstruction and more or less pronounced circulatory disturbances. On the other hand, the course of the chronic intussusception form is marked by a smaller amount of circulatory disturbances and a partial or transitory total occlusion. It is important to differentiate the permanent chronic intussusception from the chronically recurrent form in which the intussusception is entirely reduced between the attacks. The chronic intussusception type differs in frequency in different age groups. Its incidence rises with age. The low frequency of chronic intussusceptions in childhood is explained by the fact that a chronic intussusception can only occur when the lumen of the intestine is wide enough to preclude its complete occlusion. However, in a newborn child the large intestine is but a few millimeters wider than the small intestine and at the age of 15 years is 2,5-3 times as wide. Even in the colon of small children intussusception is always accompanied by occlusion.
According to most publications, about one quarter of all intussusceptions are presented within 12 hours after the onset of symptoms, about 50% within 24 hours and about three quarter within 48 hours (10, 156, 234, 236, 263, 336, 344, 375). But there is also a number of reports saying that about half the cases are presented within 12 hours, two thirds within 24 hours and almost 90% within 2 days (17, 62, 110, 129, 332). Chronic intussusceptions are uncommon in children (2-10%) (44, 200, 276), but more common in adults (about 60%) (227, 228, 334).

II.4. EPIDEMIOLOGY

II.4.1. Incidence

Intussusceptions have been reported to form 5 to 20% of all intestinal obstructions at all ages (249, 259). 80 to 90% of intestinal obstruction in infants are due to intussusception. Intussusception is the most important cause of intestinal obstruction between the age of 3 months and 5 years (259).

The occurrence of intussusception is variable. Significant geographic variations exist between countries and regions. The reported incidence of acute intussusception is 1.5 per 1000 live births in Australia and England (202, 315, 317). In Aberdeen, Gothenburg and New York an incidence of just over 2 per 1000 live births has been calculated (24, 32, 329). In Scotland, an incidence of 2.8 per 1000 live births has been found (62, 156, 256). The incidence of intussusception in Scotland seems to have declined over recent years (257). The highest incidence has been calculated in Newcastle, 3.8 and 4.3 per 1000 live births (56, 324). A report from New Zealand mentions an incidence of 0.64 intussusceptions per 1000 live births (270). The incidence in the Netherlands that can be calculated from numbers of the SIG (Stichting Informatiecentrum voor de Gezondheidszorg), is 1.3 per 1000 live births (330).

II.4.2. Geographical variation

Geographical variation in the presentation of diseases is well documented but rarely as clearly as with intussusceptions. Intussusception in the tropics shows features that differentiate it from intussusception in temperate countries. In reports from Africa (49, 170) and Asia (8, 64, 269) intussusception seems to be a more common disease, especially in older children and adults. At the same time the incidence in infants seems to be lower in the tropics. Besides a male preponderance, a subacute or chronic course with mild symptoms seems to be characteristic in tropical intussusception. The percentage of idiopathic non-infantile intussusceptions around the ileocecal boundary is high compared to temperate climates. Amebiasis, abdominal tuberculosis and ascaris infestation form a diagnostic problem and might play an etiological role in tropical intussusceptions. On the other hand intussusception with a more Western-like pattern has been described in reports from Ethiopia (357) and Jamaica (312).

II.4.3. Seasonal incidence

A marked seasonal variation in incidence of intussusception has been suggested in many reports. Unfortunately these reports are contradictory. Some authors calculated a higher incidence in late spring and summer (32, 75, 164, 261, 262, 270, 323). Others reported a peak incidence in winter and early spring (9, 174, 181, 257, 332). Clarke found the highest incidence in spring (45). Nyborg found two thirds of all intussusceptions in the first 5 months of the year (239). Several reports describe two or more peaks (24, 49, 170, 281, 32). A markedly lower incidence during winter (170, 315) or

autumn (262) has been reported. It is not surprising that many authors found no significant seasonal distribution (62, 156, 225, 231, 336). Pollet stressed that season had a marked effect on incidence among children over one year old. He had no explanation for the statistically significant low figures in infants and the surplus in children over one year of age in the winter season (257). There is so much variation in the reported series that probably season has no significant influence. An explanation for marked monthly incidence has been sought in a correlation between the seasonal peaks of respiratory tract infections, gastroenteritis and intussusception. This corresponds to theories concerning the etiological role of viruses in intussusception. Publications in which no correlation between the incidence peaks was found (32, 49, 75, 225, 239, 312, 315) outnumber those in which such a correlation was established (332). Potter found a correlation with respiratory tract infections but not with gastroenteritis (261). Other explanations for peaks in the incidence of intussusception given in the literature are the addition of fresh vegetables to the child's food in the month of May and errors in diet which are apt to occur at the Christmas and New Year festivities.

II.4.4. Distribution according to gender

All series report a strong male preponderance, the great majority reporting a male-female distribution of 3:2 to 3:1. It has been stated that this male preponderance is most pronounced in infancy (370). In other publications (89, 164, 179, 239, 323) an even stronger male preponderance over the age of 1 year was found. In intussusceptions in babies under 4 months of age no preponderance was found (230). In reports on intussusceptions in children over 2 years of age (89, 92, 308, 350), about the same well-known distribution was found. In adult intussusception, there appears to be no predilection for either sex (3, 228, 366) with both male (65) and female preponderance (290, 334) reported in various studies.

There is no plausible explanation for the male preponderance. Kock (1913) was able to demonstrate that a strong pressure against the spermatic cord of a cat caused contraction of the caecum. He believed that the same effect caused by pressure from screaming, hydrocele or inguinal hernia in a child would give rise to a spastic contraction in the caecum with an intussusception as a result. Obadalek (1929) tried to produce such a contraction at laparotomies by pulling the boys testicles but without success. In Gibson's opinion, increased abdominal pressure caused by a phimosis, produces intussusception. Laurell (1932) pointed out that a transitory incarceration of a part of the caecum in an open processus

vaginalis results in swelling of the caecal wall. This swelling would cause intussusception (164, 179, 239, 240).

II.4.5. Racial incidence

Apart from the above mentioned geographical differences, studies of the absolute incidence of intussusception in different races in one country are not available. Intussusception in the US may be more common in white children (271, 281) or in black children (1). At most the difference is not great.

II.4.6. Heredity

The coincidental occurrence of intussusception in parent and offspring (343), in twins (342), in siblings (164, 179, 202) and relatives (143) has been described. However there is no evidence that there is any increased likelihood of intussusception in a sibling once a child in a family has been affected. According to Ravitch a surprising number of children with intussusception occupies late positions in their mothers obstetrical careers (272). In contradistinction, several authors found a high percentage (47-64%) of firstborn children among their patients (164, 240). MacMahon found no consistent trend with birth order (202).

II.4.7. Distribution according to age

Intussusception is a typical childhood disease. Although it occurs in all age groups from newborns to adolescents, it is reported primarily in children under 1 year of age. In most series a percentage for infants between 55 and 70 has been reported (1, 20, 62, 68, 75, 156, 174, 216, 236, 281, 315, 332, 337) although many authors mention slightly higher (10, 17, 270, 323, 336, 375) or slightly lower percentages (11, 32, 234, 239, 259, 362). It is striking that in three publications from Ireland a very high percentage of infants is reported, between 81 to 94% (57, 196, 263). This may point to a geographical variation. Almost just as striking is the fact that 3 of 5 series reporting rather low percentages of infants, 33 to 41%, are from Sweden and Danmark (24, 44, 110, 129, 291).
Children under 2 years of age form 70 to 85% of all pediatric cases (1, 11, 17, 20, 62, 68, 75, 156, 174, 234, 236, 239, 253, 259, 270, 281, 315, 323, 332, 362). Authors who report a markedly higher or lower percentage of infants also found a higher or lower percentage for children under the age of 2 years (24, 32, 44, 57, 110, 129, 196, 263, 291).

Children over the age of 4 years account for about 15% of all cases (24, 174, 234, 239, 315, 332, 362). Little higher percentages (about 20%) have been given by Gierup and Abbott (1, 110). Much lower numbers were found by Auldist, Minami, Wright and Swenson (10, 216, 337, 375).
The reported mean age varies greatly from 6.5 months (196) to 24 months (32, 256). The peak period for intussusceptions lies between the third and ninth month (1, 10, 24, 57, 75, 156, 162, 174, 216, 239, 256, 270281, 337, 375). Intussusception in the first month of life is rare.

II.4.7.1. Intrauterine intussusception

Experimental and clinical (198) evidence has been obtained that vascular accidents account for part of intestinal atresia's, especially small intestinal atresia's. Intrauterine intussusception as a cause of intestinal atresia was mentioned by Chiari in 1888. Atresia caused by intrauterine intussusception varies from 0.6 to 13.1% of all atresia's (132, 345). The clinical picture of a newborn child with atresia due to intrauterine intussusception is characterized by vomiting and abdominal distension (245, 247) and often the passing of normal meconium in which lanugo hair and squamous cells can be detected (245, 345). Sometimes necrotic tissue is lost per anum (120). Signs of a previous meconium peritonitis or ascites can be found (245). Plain abdominal radiographs show signs of a mid-intestinal obstruction. Barium enema in cases of mid or low ileal obstruction due to atresia as well as meconium ileus can show a 'microcolon' as a reflection of the shutting off of the colon (even though it may be hours or days before birth) from the flow of meconium and succus entericus. On the other hand, in an important proportion of cases, a normal-sized or nearly normal-sized colon has been observed (345). In many reports evidence has been presented of intrauterine intussusception very late in the course of gestation. In these cases the differential diagnosis with neonatal intussusception may be very difficult. All these children are full-term babies, with small intestinal atresia and without any associated anomalies (345, 120). In contradistinction children with atresia due to volvulus or internal hernia are premature in 35% of cases (120).
At operation, an atresia with two connected or disconnected blind ends with or without a corresponding gap in the mesentery, is always found with significant dilatation of the proximal intestinal segment. Generally, a plug can be felt in the most distal segment (148, 245, 345). Histological examination, macro- and microscopically, reveals remnants of the intussusception. Three types can be

found: complete intussusceptum (Figure 12a), polypoid mass containing bowel tissue, not always in direct contact with the proximal ending of the distal segment (Figure 12b), or microscopical remnants of bowel tissue located in the wall of the distal segment (Figure 12c) (345, 132). There has been speculation concerning the cause of intrauterine intussusception. Louw et al. thought atresia to be the cause but this fails to explain intrauterine intussusception without atresia or gangrene (198, 148). Another cause stipulated is viscid meconium that might stimulate the fetal intestine to excessive peristalsis. Ileal atresia is found in 15-20% in combination with meconium ileus but the combination of ileal atresia due to intrauterine intussusception with mucoviscidosis has never been reported (120). The survival rate after surgical correction of atresia with intrauterine intussusception is 80% (345).

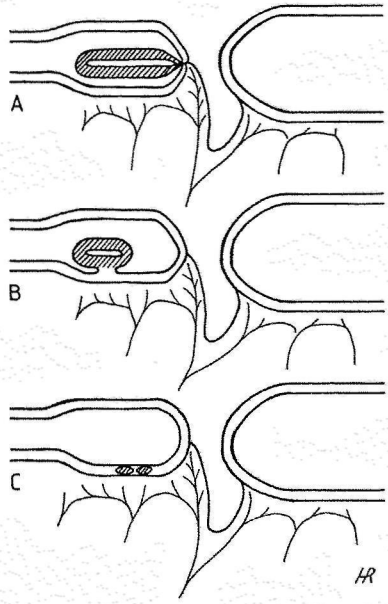

Fig. 12a,b and c. Three histologic types of atresia due to intra-uterine intussusception: A: Complete intussusceptum, B: Polypoid mass of bowel tissue, C: Microscopical remnants of bowel tissue.

II.4.7.2. Intussusception in neonates

Intussusception during the first month of life is much less common than in older infants (246, 338). An incidence of 0.3 to 0.8% of all intussusceptions has been reported (32, 281, 265). The clinical features of neonatal intussusception differ strikingly from that in older infants and children. Colicky pain (378) and an abdominal

mass are often missing in the neonatal period. The major symptoms are vomiting and abdominal distension indicating bowel obstruction. Rectal bleeding is also an important but late sign (246, 338, 378). Pathological leading points such as Meckel's diverticulum, duplication cyst (244, 246), polyps (265, 338) and hamartoma are present in about one third of cases (246, 378). Small intestinal intussusceptions are frequently found (113, 318, 338, 378). These factors limit the therapeutic value of gastrografin enemas but successful attempts have been reported (166). The diagnosis can be made by ultrasonography as well (244). Most of the times the treatment must be surgical (378). There still is a significant morbidity and mortality (318, 338, 378), especially in those cases occurring in prematures (113, 318, 331). The differential diagnosis with necrotizing enterocolitis (318, 331) and meconium obstruction disease (113, 326) form a diagnostic challenge. Combinations have been seen.

II.5. INTUSSUSCEPTION IN OTHER SPECIES

Intussusception has been reported in all kinds of animals, mammals and birds. Most reports concern single cases. Intussusception in a monkey (Papio papio) has been related to candidosis. An idiopathic intussusception was found in a Galago senegalensis. A dramatic intussusception due to a fishhook has been described in a Florida manatee (sea cow). Ileocecal intussusception in young fowls has been associated with starvation and with ox blood meals . In young rabbits ileoileal intussusceptions in combination with intestinal coccidiosis (Eimeria perforans) have been reported. Intussusception has been a complication in laboratory rodents. In a group of 77 young guinea-pigs, which died shortly after weaning, 49 small intestinal intussusceptions were found. Mortality normalized after withdrawal of lucerne hay as a food, possibly containing G1 aflatoxin (307). Over a period of 11 months six cases of colonic intussusception were described in a group 120 young golden hamsters (Mesocricetus auratus) receiving intragastric inoculations of live Salmonella organisms as part of a study on the mechanisms of food poisoning (258). Coggin et al. developed an animal model for spontaneous lymphoma induction by an infectious DNA viroid-like agent in hamsters (Mesocricetus auratus). Tumour growth within the gut wall, arising within Peyer's patches of the small intestine was noted. Massive enterocolic and colocolic intussusceptions occurred in primarily young hamsters within 45 days of exposure. No incidence was given (47). During control tests on vaccines containing Bordetella pertussis cells preserved with Thiomersal unexpected deaths in young mice have been encountered mostly due to ileocecal

intussusceptions. The incidence was up to 40% (25, 26). In the biochemical literature evidence can be found that pertussis toxin can play an important role in the communication at the level of the cell membrane (135, 376). A high incidence of intussusception has been found in a single family line of mice with a mutant colour gene through X-ray experimentation (145). Intussusception in mated female mice in combination with nematode infestation and focal liver necrosis has been reported (226). Intussusceptions in cats are mainly described in kittens and are associated with constipation, abdominal straining, close interbreeding, foods and foreign bodies. Foreign bodies are the far most common cause of intestinal obstruction in dogs and seven times as frequent as the second cause, intussusception (180). Intussusception in dogs has a predilection for the younger age, a tendency to recur within 20 days and a high need for resection. No predisposing factors other than diarrhoea associated with transfer from a local dog home to private ownership, have been mentioned as a contributory factor (233, 364). Niemand mentions thymus dysfunction as an etiologic factor (233). Gastro-esophageal intussusception including spleen, duodenum, omentum and pancreas in puppies and older dogs is reported. A significant number of postoperative intussusceptions after renal transplantations in adult mongrel dogs (12/38, 10/138) and after pancreas transplantations in puppies (5/30) has been described. To prevent this complication the administration of an anticholinergic drug has been advocated (73, 242). Others found a marked decrease of the number of postoperative intussusceptions after intra-operative administration of morphine (210). Houck reported 11 intussusceptions in dogs after bilateral nephrectomy, all within 16 days after operation with one exception (35 days) (150).
All reports on intussusception in cattle and horses concern cases. Intussusception in the bovine is uncommon. Successful resections have been described. Intussusception in horses occurs primarily in younger animals. An important proportion is of the ceco-cecal, ceco-colic or colo-colic type. Resection is necessary. Tapeworm lesions may play an etiological role (5, 13).
Reading these reports one gets the impression that intussusception in other species is also a disease in younger specimens, primarily located at the ileocecal boundary and where infectious agents might play a role in the pathogenesis.

II.6. ETIOLOGY OF INTUSSUSCEPTION

The etiology of most intussusceptions is not understood. The majority of cases is called idiopathic. Many theories have been advanced

in an attempt to explain causative factors involved in idiopathic intussusception. Numerous etiologic factors have been described and this often illustrates how little is known for certain. Several authors state that an intussusception arises from the interaction of 2 or more factors (30, 237, 239). This is an explanation for the fact that most etiologic factors have a far more higher incidence than intussusception caused by these factors. All factors proposed in the literature can be subdivided in the following way:

- Factors related to the contraction state of the bowel (the paralytic intussusception theory, the spastic intussusception theory, dysfunction of the vegetative nervous system).

- Factors related to local anatomic characteristics.

- Local mechanical factors, so called leading points inclusive.

- Factors related to infection (viral, bacterial, parasitic) and to inflammation.

- Miscellaneous factors (intestinal intubation, postoperative, pregnancy, trauma).

II.6.1. Factors related to the contraction state of the bowel

II.6.1.1. Paralysis

As early as 1677 Peyer attempted to explain the origin of intussusception by assuming that a paralysed intestinal part drew an adjacent, contracted portion into itself. Throughout centuries this paralytic intussusception theory had its advocates (164, 239).

II.6.1.2. Spasm

In the eighteenth century Rigall thought a spasm to be the beginning of an intussusception (287). The spastic intussusception theory assumed a limited intestinal spasm could be brought into the adjacent lumen of the distal intestinal loop by means of peristalsis of the intestinal portion situated above it.
Nothnagel who supported this theory, showed in his experiments on rabbit gut that a local spasm, produced by stimulation with faradaic current, could give rise to intussusception. He showed that the contracted intestinal part was also prolonged so that at both ends the adjacent intestine was slightly overlapping the contracted portion. The musculature of the part of the intestinal wall, encom-

passed in this way, was stimulated to peristalsis after a brief interval by the intruded intestinal portion. This resulted in the development of an intussusception in an anal direction while, on the other hand, the oral encompassment was reduced (237). However the cause of the spasm remained unknown in most (idiopathic) cases. Stalked and stalkless tumours may cause a spasm. They form a disturbing element in the peristalsis which, in the end, amounts to an irritation, producing the primary spasm which constitutes the necessary prerequisite for the origination of an intussusception. The intussusception tumour acts as a polyp and stimulates to a lively peristalsis. In this way a double intussusception may arise. Inflammations in the intestinal mucous membrane are cited as a common cause of hyperperistalsis and of the primary spasm. This contraction is caused in either of two ways, directly, by lowering the irritation threshold of the intestine, or, indirectly, via a lymphatic swelling originating in the inflammation (239). Spasms can also be traumatic or psychologic in origin or they are caused by the pressure from the outside by a tumour, for instance in pregnancy. Hyperperistalsis due to bowel obstruction e.i. volvulus, internal herniation can cause spasm.
Recently Rokitsky and Morozov claimed that functional disorders of the gastrointestinal tract in young children are mostly consequent upon birth trauma of the cervical spine and spinal arteries. This was confirmed by the observation of experimental trauma to the cervical portion of the vertebral column in which stable spasm of the small intestine occurred and the large intestine remained functionally intact (289). Kock caused contraction of the caecum in the cat by strong pressure against the funicle (179). Obadalek failed to produce such a contraction by pulling boys testicles at laparotomy (240).
Antiperistalsis in the small intestine is supposed to work anti-intussusceptogenic. Antiperistalsis in the large intestine possibly plays an important role in the origination of intussusceptions around the ileocecal valve. There is much more antiperistalsis in the colon. It causes the bowel contents to stay longer at the same place so there is more time for the resorption of water. Peristalsis in the first part of the colon pushes a protruding valvula Bauhini further into the colon ascendens while anti-peristalsis pushes it further into the caecum.

II.6.1.3. Autonomic nervous system dysfunction

Another etiologic factor in the origin of intussusceptions may be a dysfunction of the nerve control, i.e. of the autonomous nervous system. Some assume the absence of parasympathetic innervation

to the large intestine but this assumption seems incorrect as a stimulation of the vagus brings about a contraction of the proximal colon and a stimulation of the pelvic nerve gives a contraction in the distal colon. Others advance a dysbalance in favour of the sympathetic component of the autonomous nervous system. A retardation of the peristaltic movements in the intestinal canal and an increased sphincter tonus result in a strong dilatation. Nyborg believes that in patients with intussusceptions the para-sympathetic component is stronger. This has an insufficient valvula Bauhini as a consequence, bringing about an ileum prolapse. De Jong states that a decreased function of the thymus results in a strong para-sympathetic influence on the small intestine (164). Severe spasms result. A high incidence of intussusception (30%) after resection of the thymus in dogs has been reported. Dysfunction of the autonomic nervous system and mural neural structures caused by anoxia and asynchronous neuromuscular development may also disturb intestinal contraction (164, 239).

II.6.2. Factors related to local anatomic characteristics

Since the intussusception disease is most frequent in childhood, attention has for a long time been directed to certain factors of predisposition within the intestinal canal of a child. A child's mesentery is considerably longer than that of an adult in proportion to the length of the body, and the mesentery is deficient in fat. At the time of birth, the large intestine is only a few millimeters wider than the small intestine while, at the age of 15 years, the width is 2,5-3 times as large. So, at infancy, the spontaneous reduction of an intussusception is less likely to occur.
Insufficiency of the valvula Bauhini as a result from the rapid growth of the large intestine, in the first 6 months of life, might allow a piece of the terminal ileum to prolapse in the colon, 45 per cent of all children possess a mobile caecum as against 17 per cent of all adults. Also the thinness of the intestinal wall in a child, is called predisposing. A more pronounced irritability in the intestinal canal of children as against that of adults, has been emphasized as a causative factor. The intestine of a child is more untrained than that of an adult and will react more strongly to an irritating substance. In support of this view regarding the great significance of the lymphoid tissue Perrin pointed out that Peyer's patches, accumulated in the ileocecal region, will increase very rapidly during the second half of the first year of life (249).
In the normal development of the intestinal tract the ileocecal valve protrudes into the caecum during the first year of life. Abnormally far protrusion causes intussusception. Other local anatomic charac-

teristics, not associated with age, may play an etiological role.
The longitudinal muscle layer of the intestine decreases in volume toward the cecum and is lacking at some places entirely (164, 305). Not spanning across the base of the valve commissure of the longitudinal muscle fibers has been described in connection with intussusception. Scheye thought the lesser resistance of the cecal wall and the papillary type of valve to be intussusceptogenic (305). The angle between the terminal ileum and the colon varies greatly. This angle can be 90 degrees but also almost 180 degrees, the ileum in a direct line with the colon and more prone to invaginate. Significant individual variations of the lumen of the valvula Bauhini have been described. Brown described a mesenteric vascular band, the extension of the ileocecal mesentery across the ileocecal valve (30). She presumed this band to be of etiologic importance in combination with enlarged lymph nodes in the ileocolic fossa and ileocolic fold, and together with fat accumulation in the mesentery of the well-nourished child.

II.6.2.1. Appendiceal intussusception

The incidence of appendiceal intussusception has been found to be 0.01% in a study of 71.000 appendectomy specimens (50). There definitely is a predilection age, the majority of cases occurring in the first decade of life. It is seen four times more frequently in males than in females. However in a review of 54 cases of adult appendiceal intussusception no such male preponderance was found. Four possible clinical pictures have been described: 1. acute appendicitis, 2. acute intussusception, 3. chronic intussusception and 4. asymptomatic cases. From an etiologic viewpoint the following anatomical factors are important: a fetal type of cecum, a funnel-shaped, nonfixed appendix capable of peristalsis and a thin mesoappendix. Etiologically important conditions causing irritation leading to increased peristalsis, may be intraluminal (foreign body, fecalith, worm) or intramural (mucocele, tumour, lymphoid follicle, endometrioma) (101, 190).
Most authors agree on the fact that primary resection is not the therapy of choice. A more conservative approach seems justified as only a small minority of all cases of adult appendiceal intussusception is associated with a malignancy. In case a caecal mass turns out to be an intussuscepted appendix, and at inspection no sign of malignancy is found, an attempt at reduction is made. If reduction succeeds, simple appendectomy should be performed. If reduction of the appendix fails, the caecum is opened after the mesoappendix is ligated. Then the appendix is excised with a rim of caecum. If there is a suggestion of neoplasm, frozen section should be performed.

The presence of a malignancy of the appendix is an indication for right hemicolectomy (190).
Barium enema, ultrasonography and colonoscopy can be of diagnostic significance. However, endoscopic appendectomy is not recommended and can lead to peritonitis up to three days after removal. If performed due to initial misdiagnosis, immediate surgical intervention is indicated (95).
Inversion of the whole appendix has been proposed as part of the treatment of acute appendicitis. Natural appendiceal intussusception leads in 50% to more compound cecocolic intussusception. This risk forms an absolute contraindication for this treatment, besides the fact that it may cause confusion in interpreting a barium radiography. Although rarely, even an appendiceal stump after appendectomy may lead to intussusception. In these cases cecopexy may be adequate (232).

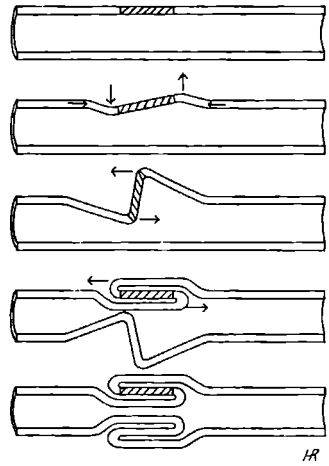

Fig. 13. Local inhomogeneity in the bowel wall precipitating intussusception.

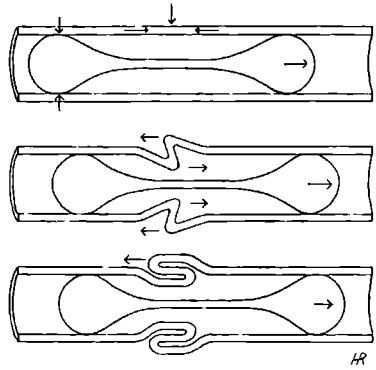

Fig. 14. Mechanical linkage of nonadjacent bowel segments precipitating intussusception.

II.6.3. Local mechanical factors

Reymond attempted to explain from a theoretical analysis the mechanism of 'kink' formation in the bowel wall which he thought to be the precursor to intussusception. He categorized the etiological factors of intussusception into two common denominators. He distinguished a local inhomogeneity in the bowel wall and a mechanical linkage of nonadjacent bowel segments (Figure 13 and 14). Among the inhomogeneities he differentiated indurated lesions, flaccid lesions and sudden changes in caliber of the bowel. Under the influence of the contracting longitudinal muscle fibers, the bowel begins to shorten, except in the inhomogeneity. The inhomogeneity can be perturbed by a small displacement, such as secondary to a change in the tone of the adjacent circular muscle fibers. A couple arises and a resultant torque tends to rotate the inhomogeneity. An intussusception may be the result.
There are intraluminal and extraluminal linkages. The forces exerted on mechanical linkages by the bowel segments are generated by peristaltic activity. The intervening bowel wall between the segments comes under compression. This system can be perturbed by a change in the tone of the circular muscle fibers. The lines of the acting force no longer coincide, resulting in a torque and in kink formation. An intussusception may be the result (285, 286).
Indurated lesions forming local inhomogeneities of the bowel wall consist mainly of bowel lesions also known as leading points (German: Leitgebilde). The proportion of intussusceptions caused by leading points varies with age of the patient, geographical region, type of intussusception. Patients with intussusception due to a leading point are older than patients with idiopathic intussusception (37, 77, 167, 264). These children are more severely ill (77). Pain, an abdominal mass and rectal bleeding are less frequent and the delay in diagnosis and treatment is longer than in idiopathic intussusception (37, 77, 167). These intussusceptions are unlikely to be reduced by hydrostatic barium enema and will almost certainly require surgical reduction (77, 264). However, successful attempts at hydrostatic reduction of these intussusceptions have been reported (82). At laparotomy the intussusception is found to be of the enteric type and irreducible or gangrenous more frequently than in idiopathic intussusceptions. So, often resection is required. Commonly a high complication rate is found (37, 77.167, 264).

II.6.3.1. Meckel's diverticulum

Meckel's diverticula account for more than 50% of all leading points (11, 32, 62, 77, 110, 156, 167, 332, 362) in children. A Meckel's

diverticulum is present in approximately 2% of the population (177, 360), but only 4% of these become symptomatic. Between 4 and 14% of the complications of Meckel's diverticulum can be attributed to intussusception. A Meckel's diverticulum is a factor in about 2,5% of intussusceptions (130). It appears that persons with a Meckel's diverticulum may not be more likely to develop intussusception than normal individuals. The Meckel's diverticulum acts as a local inhomogeneity when it adheres with fibrin to the serosa of the adjacent bowel. On the other hand the congenital mesodiverticular band of the diverticulum may act as a disturbance for the peristalsis. A Meckel's diverticulum may also invert itself into the bowel lumen and have the character of a link of non-adjacent bowel segments (285, 286). This invagination may be facilitated by an intramural tumour, ectopic gastric or pancreatic tissue. The diverticulum is not necessarily found at the apex of the intussusceptum (239). Non-Meckelian congenital or adult small intestinal diverticula may cause intussusception acting in the same way (320).

II.6.3.2. Cysts

Enterogenous and duplication cysts may be incorporated in the wall of the normal intestinal tract or in the mesentery (221, 333, 360). 60% of all duplication cysts are localized in the small intestine (299) and 40% in the ileum (252). Duplication and enterogenous cysts account for about 5% of leading points in childhood intussusception (11, 32, 37, 62, 69, 77, 82, 110, 122, 156, 167, 252, 264, 281, 332, 336, 362). These cysts form a local inhomogeneity of the intestinal wall. Gross reported 2 duplication cysts giving rise to intussusception out of a series of 64 cysts (122). Grosfeld reported on 23 duplication cysts. None of them presented as an intussusception (121).

II.6.3.3. Heterotopic tissue

A nodule of heterotopic tissue may also act as an inhomogeneity of the bowel wall. Small asymptomatic gastric heterotopias are relatively common but large, clinically significant cases are rare. Their incidence in the duodenum is highest. They may give rise to obstruction (often intussusception), peptic ulceration or haemorrhage. Gastric heterotopias are acquired or congential. Acquired gastric heterotopia is associated with various inflammatory disorders. The combination of intussusception and congenital heterotopic gastric or pancreatic tissue is almost always associated with intestinal duplication, Meckel's diverticulum or diverticula of an other origin. Heterotopic gastric or pancreatic tissue not associated with other

anomalies is a rare leading point (167). Large series of intussusception fail to report it as an etiologic factor (32, 62, 68, 69, 75, 77, 110, 156, 332, 336, 362). No instance of intussusception was reported in a large series of aberrant pancreatic tissue as the sole abnormality.

II.6.3.4. Benign tumors

The minority of benign intestinal tumours causes symptoms, such as intestinal obstruction and anemia due to bleeding. Intussusception is not uncommon. Benign tumours, most of the times polyps, account for about 20% of all leading points in childhood intussusceptions (11, 32, 37, 62, 68, 69, 75, 110, 156, 167, 264, 281, 332, 336, 362). The major part is located in the small intestine. The gastroduodenal type has been described (229). Tumours, especially stalked tumours, stimulate invagination partly because of their weight, partly owing to the fact that they are involved in the peristalsis, thus occasioning a lateral intussusception which, after some further pulling, is transformed into a central one. Yet, also small, stalkless tumours may cause a spasm. Most of the tumours causing intussusception are adenomatous polyps (175, 229, 264). These arise from the superficial mucus membrane or intestinal glands. An important proportion exists of inflammatory fibroid polyps, a group of nonspecific submucous granulomatous lesions of unknown etiology with eosinophilic infiltration. Hamartomas form another important group. These are localized overgrowths of normal mature cells identical to the types constituting the organ in which the tumour is found. Within this group the Peutz-Jeghers syndrome has a special place. This dominant autosomal syndrome, characterized by intestinal polyposis and mucocutaneous pigmentations is associated with chronically recurrent intussusception (177, 221, 333, 348). Most submucous lipomas do not cause symptoms but if they do, an intussusception is common (in up to 73%) (53, 163). Leiomyomas (74), haemangiomas (221, 264) and neurofibromas (221) are rare leading points of intussusception.

II.6.3.5. Malignant tumors

Intussusception as a complication of malignant neoplasm is common in adults (30%) (65, 290) but rare in children (0.5%) (32, 37, 62, 68 75, 77, 110, 156, 167, 264, 281, 332, 336, 362). Adenocarcinoma is the most frequent primary tumour acting as a leading point in adult intussusception (169). 5-10% of all caecal adenocarcinomas in adults present as intussusception (65, 290). Other, rare primary malignancies as plasmacytoma, leiomyosarcoma and car-

cinoid have been reported as leading point in adult intussusception. Most of these intussusceptions occur at or beyond the cecal valve but the gastroduodenal type also has been reported. Primary resection is indicated.
Just as important are metastatic tumours. Usually, they give rise to obstruction and chronic intussusception of the jejunum. Primary tumours are malignant melanoma, carcinoma of the lung, carcinoma of the breast, carcinoma of the ovary, carcinoma of the kidney, leukemia, carcinoma of the stomach and fibrosarcoma of the buttock (41, 169). If the general condition of the patient is good, treatment must be surgical. An attempt at manual reduction is justified and will not influence the patients prognosis. Often the resection can be limited. Idiopathic intussusception in adults with disseminated malignancy has been reported (169). Recurrence from other metastases must be kept in mind.
Primary lymphosarcoma of the gastrointestinal tract account for about 2% of all malignant tumours in childhood (161). Most of them are located in the ileum. They present as an intussusception in about 40% (161, 213). The great majority of these intussusceptions occur in children between 3 and 13 years of age. The course of the intussusception is almost always chronic and the location around the ileocecal valve with the lymphosarcoma in the terminal ileum (81, 169, 363). Lymphosarcoma accounts for about 7% of all leading points of intussusception (11, 32, 37, 62, 68, 69, 75, 77, 110, 156, 167, 264, 281, 332, 336, 362). One or two cases are mentioned in most recent, large series. Hydrostatic reduction of intussusception caused by a lymphosarcoma is rare but has been reported (82). Careful observation of postreduction X-rays combined with upper gastrointestinal series has been advocated in older children (82, 363).
Intussusception due to other malignancies is very rare. Fibrosarcoma, leiomyosarcoma and metastases of rhabdomyosarcoma and of lymphosarcoma as leading point have been reported (69, 70, 264).

II.6.3.6. Henoch-Schönlein purpura

Henoch-Schönlein purpura is a non-thrombocytopenic purpura related to increased vascular fragility with mononuclear perivascular cellular infiltrates and thrombi. This effects skin, mucous membranes, kidneys, joints and the wall of the intestinal tract (29). Its origin might be immunologic. It is mainly a childhood disease (4-15 years) (84). Symptoms are a rash, urticaria, pain, often abdominal with vomiting, diarrhoea and rectal blood loss (18, 29). Subserosal and submucosal haemorrhages with edema which may cause obstruction and gangrene, may also increase peristalsis and act as

a leading point of intussusception (18, 91). This happens in about 5% of all children with Henoch-Schönlein purpura (18, 177, 206). In many large series 1 or 2 of these cases are reported (32, 68, 69, 75, 156, 167, 332). It accounts for about 7% of all leading points. Suita in a series of 485 intussusceptions encountered even 11 cases of Henoch-Schönlein purpura among 29 leading points. The main part (65%) of these intussusceptions is enteric (91, 206). The differential diagnosis between uncomplicated Henoch-Schönlein purpura and purpura complicated by intussusception is difficult, clinically but also with the aid of barium enema. Only 25% occurs around the ileocecal valve. Vomiting, diarrhoea, rectal blood loss and crampy abdominal pain is present in 50% of uncomplicated cases and may precede the purpura. Peritoneal signs and a marked leucocytosis must be alarming (18). The use of ultrasonography may prove a very helpful, non-invasive, low cost and easy to repeat diagnostic aid (206). Thrombopenic diseases and coagulation disorders may cause intussusception by submucosal hematoma formation but are rarely reported.

II.6.3.7. Mucoviscidosis

Mucoviscidosis or cystic fibrosis is a disorder of exocrine glands with the production of either an abnormal glycoprotein more viscid than normal mucin or a variation in polymerization of normal mucin. Mucoviscidosis is inherited on an autosomal recessive basis (221). Stools often consists of a thick putty-like material that is adherent to the intestinal wall over a long distance. This may give rise to an inhomogeneity of the wall but also to mechanical linkage of non-adjacent bowel segments, and cause intussusception. All children with mucoviscidosis and intussusception reported in the literature were over the age of 4 years. The incidence varies between 0.2 and 1% which is high for this age group. Usually the course of these intussusceptions is acute and the localization is around the ileocecal valve. The differential diagnosis with uncomplicated mucoviscidosis (often causing abdominal pain) and meconium ileus can be problematic. The occurrence of intussusception has no relation to the effect of treatment of mucoviscidosis (146, 372).

II.6.3.8. Bezoars

The term 'bezoar' refers to concretions of various types found in the gastrointestinal tract of man and animals. Bezoars can be of various material like hair, wool, stone, cellulose, resinplants.
Bezoars can be very long, extending from the stomach to the terminal ileum, multiple or connected to a daughter bezoar. The

commonest complication is intestinal obstruction although gastroduodenal ulceration with haemorrhage or perforation may occur. By acting as a linkage between non-adjacent bowel segments, as meant by Reymond (285, 286), a bezoar can cause an intussusception. Wherever the site of the obstruction, a thorough search must be made at laparotomy to exclude multiple concretions. This dictum is often forgotten and has led to many re-operations in the past (114, 283).

II.6.4. Factors related to infection and inflammation

II.6.4.1. Viral infection

The infection theory is one if not the most predominant explanation for the origin of intussusception. This hypothesis accounts for both facts that intussusception takes place mainly before the end of the first year of life and that in most cases it starts at or near the ileocecal valve. Perrin pointed out that the amount of lymphoid tissue i.e. Peyers plaques, at the ileocecal valve and in the terminal ileum, was greatest up to one year of age (249). Hyperplasia of this tissue could make it act as a leading point (194). There was evidence that infection could produce hyperplasia of submucous lymphoid tissue and of mesenteric lymphnodes.
It was suggested that seasonal peaks in the incidence of intussusception might be associated with seasonal increases in enteric and respiratory infections (249, 276). Although no significant seasonal preponderance has been found, the peaks of intussusception and respiratory tract infections have been reported to coincide.
Viruses of the adenoidal (A)- pharyngeal (P)- conjunctival(C) group were isolated from the mesenteric glands and stool of cases of non-specific mesenteric adenitis (293). In upper respiratory infections these viruses were easily isolated from the stools. In november 1959 Rutten presented 3 cases of intussusception in children associated with adenovirus infection (296, 297, 298). In the sixties and seventies controlled virologic studies of intussusception were performed in three important clinics in England (19, 107, 108, 260, 261, 262, 292, 293). In broad outline their results were the same. Adenovirus infection was present significantly more frequently in patients with idiopathic acute intussusception than in controls. Children with intussusception had a respiratory infection in 29 to 56% (107, 261, 292, 293). Mesenteric adenitis was often observed (292, 293). Adenovirus (type 1, 2, 5 and 6) was isolated from faeces in about 60% (107, 108, 262, 292.293). Throat swabs showed adenovirus in 19 to 48% (108, 261, 293). Adenovirus could be isolated from mesenteric glands in 33 to 65% (19, 108, 262, 293). Serological

evidence for concurrent or recent adenovirus infection, a fourfold rise in antibody levels to adenovirus (mainly type 1, 2, 5, and 6), was found in 51 to 69% (19, 108, 262, 292, 293). These findings were significant when compared with a number of control groups. They indicate that an active and probably recent systemic viral infection is present in an important proportion of acute idiopathic intussusception. The conclusion was that viruses, particularly adenoviruses, play a causal part in the etiology of intussusception.
At the same time several other important findings were reported. The incidence of past infection by 'epidemic' adenovirus, type 3 and 7, was the same in children with intussusception as in the population at large. In contradistinction, children with intussusception have significantly less evidence of past infection by 'non-epidemic' adenoviruses, serotypes 1, 2, 5 and 6 (260). In addition, the serological results in the control series indicated that adenovirus infection is common in infants (260, 292). On the other hand, Gardner found that adenovirus seemed less likely to be isolated in younger affected children than in older ones. Groups with and without evidence of viral infection showed no clear clinical differences. Neither the symptoms before the onset of the intussusception, nor the presence of enlarged lymphnodes nor the course of the illness afforded any means of differentiation (108). There is serological evidence that virus isolation from stools in intussusception is not a prolonged excretion of virus from some previous, unrelated infection (262).
There was quite a lot of criticism of these studies. In addition to the existence of several causal factors which could vary in importance in different communities, the difficulties of ascertaining the prevalence of adenovirus in a general population of the same age, especially the selection of a control group of normal children, were pointed out (108). Control groups were lacking (107), ill-defined (292), were of a different mean age (19), or consisted of children admitted for minor surgical procedures, maybe exposed to infection characteristic of a closed community, as were all children with intussusception (293). The presence of signs of respiratory infection and of gastro-enteritis was not accurately assessed in the patient groups (19, 108, 260, 261). The degree of mesenteric adenitis could not be correlated with the duration of symptoms or with laboratory evidence of virus infection. The incidence of mesenteric adenitis exceeded the incidence of positive virus isolation from lymphnodes (293). Antibody response may not be the best method for detecting a virus. The intussusception could not be ruled out as a possible cause of adenovirus infection. In conclusion, although adenovirus infection is probably capable of producing hyperperistalsis and some projection of lymphoid tissue into the lumen of the bowel, many children undergo infection by adenovirus within the first few years

of life without signs of intussusception and on the other hand many intussusceptions occur without any evidence of virus infection. In more recent studies from outside the United Kingdom a far lower incidence of adenovirus infection, proved by isolation from faeces (45, 231, 380), light and electron microscopic examination (381), throat swab and serologic evidence (231), was reported (25-39%), as low as in previous control groups. Infants with intussusception and adenovirus infection outnumbered children older than 1 year of age (45, 231, 380). The monthly distribution of cases did not indicate a seasonal preponderance (45, 380). In conclusion, in the complex etiology of intussusception the virus factor is just one among a number of contributory causes.
In the above mentioned studies also other viruses were isolated in cases of intussusception. Herpes simplex virus (107, 108, 231, 260), poliovirus (108, 231, 260), reovirus (380), ECHO type 7 and 9 (19, 231, 260, 380) and Coxsackie virus (260) were reported. Konno isolated human rotavirus from stools of children with intussusception (37%) and both electron microscopy and serologic examination gave evidence of infection with rotavirus (181, 182). Rotavirus has been established as a major etiologic agent of acute gastroenteritis, lymphoid hyperplasia and mesenteric adenitis in children in Japan. In addition, human rotavirus with intussusception occurred in the same period as epidemic gastro-enteritis caused by rotavirus. The findings of Mulcahy of Australia, however, are in every way in sharp contrast to the findings of Konno (225).

II.6.4.2. Lymphoid hyperplasia

Lymphoid tissue in the small intestine is found throughout the submucosa, most commonly presenting as aggregates of lymphoid follicles, known as Peyer's patches. Peyer's patches contain a very high number of antibody forming B-lymphocytes, suggesting an important role in the immune response. The number and size of Peyer's patches increase most rapidly during the first three years of life. The increase continues on a moderate scale through puberty. Subsequently there is a progressive decline (61). According to Fieber regression of Peyer's patches takes place after the age of 9 (96). The follicles in the small intestine increase numerically and dimensionally as the ileocecal junction is approached. In the colon they increase in number from the proximal colon down to the rectum.
Benign overgrowth of the lymphoid tissue of both large and small intestine is common in childhood. According to Louw particularly round about puberty. According to Danis its occurrence coincides with the age of increased lymphoid reactivity and it is usually seen in children less than 10 years of age (61). Nodularity secondary to

submucosal lymphatic follicles in the terminal ileum has previously been recognized as normal in the pediatric patient (102). Some call this lymphoid hyperplasia. However, it has been stated that this lymphoid hyperplasia of both small and large intestine in children is a benign situation that represents the normal hyperplasia of lymphoid tissue that occurs in childhood (4). Benign overgrowth is thought a nonspecific reactive change which may occur anywhere in the intestinal tract from the base of the tongue to the anus. The size of the follicular centres may vary with diet or various systemic infectious diseases. Lymphoid overgrowth of the small intestine has been produced in animals after injection of bacteria or irritating agents. Contrarily, diminution in lymphoid tissue in the intestinal tract and mesentery has been noted in germ-free animals (96). Other authors want to distinguish lymphoid hyperplasia as a separate clinical entity. According to them lymphoid hyperplasia represents a compensatory mechanism by the gut immune system to the repeated antigenic stimuli presented to the alimentary tract in the form of bacteria and not handled by the usual antibody response (4, 248). Lymphoid hyperplasia has been described in the literature under a variety of names including enteritis follicularis, non-sclerosing ileitis, pseudopolyposis lymphatica, ileitis catarrhalis, ileitis follicularis, gastrointestinal pseudoleukemia, nodular lymphoid hyperplasia, papillary lymphoid hyperplasia, hyperplastic lymphoid polyps and cobblestone ileum (199, 304). Lymphoid hyperplasia is mentioned in the differential diagnosis of intestinal polyposis syndromes, multiple polypoid lesions of the colon as well as the small intestine (199). Most of the time this disorder is not well defined, any increase in size of intestinal lymphoid tissue may be meant. It has been found hereditary and also in association with familial polyposis and with Gardner's syndrome. It has even been mistaken for it and treated by unwarranted total colectomy (55). In adults an interesting syndrome has been described consisting of lymphoid hyperplasia, dysgammaglobulinemia, increased susceptibility to infection, diarrhoea and lambliasis.

Lymphoid hyperplasia was not found to be associated with a specific clinical picture although rectal bleeding is a frequently associated symptom (39, 51, 55, 102). However, lymphoid hyperplasia was not the reason for rectal bleeding but one of the disorders it was found in combination with. Capitanio reported 1 case associated with ileocolic intussusception in a series of 19 children (39). In air-contrast studies of the colon mucosal nodularity and umbilication in multiple polyps appear to be diagnostic of lymphoid hyperplasia and differentiate it from juvenile and other polyps in children (39, 102). Proctoscopic examination shows small polypoid lesions crowd-

ed so closely together that the normal mucosal pattern is obliterated (51, 55). Biopsy reveals lymphoid aggregates.
Lymphoid hyperplasia of the terminal ileum is characterized by localized morphological changes of the lymphoid tissue in the intestinal (sub)mucosa, often associated with non-specific mesenteric lymphadenitis (96). At the ileocecal valve lymphoid hyperplasia of Peyer's patches has a unique circumferential disposition that could trigger the invagination of the ileum into the large intestine. Fieber proposed to distinguish lymphoid hyperplasia of the terminal ileum as a clinical entity (96). So far only Schenker considered it to be a well-circumscribed syndrome to which he added ileocolic intussusception. He described 6 cases of children with a combination of lymphoid hyperplasia of the terminal ileum and ileocolic intussusception. In addition he could state that immunodeficiency is not a part of the lymphoid hyperplasia syndrome. Serum immunoglobulines were normal in the four of six patients who were tested (304).
Perrin and Lindsay surveyed a series of 400 intussusceptions in infants and children under two years of age with almost exclusive involvement of the ileocecal region, the majority of which was caused by inflammatory swelling of the lymph tissue in the terminal 15 cm of the ileum (249). Since Perrin there have been few reports in which lymphoid hyperplasia was observed to be the actual cause of intussusception (54, 61, 83, 127, 301, 304). The lymphoid hyperplasia in most reports is merely mentioned as one of the lesions uncommonly encountered (281, 20). It has been postulated that this hyperplasia may often remain unsuspected because hyperplasia of Peyer's patches of the terminal ileum is rarely if ever demonstrable by palpation and barium enema. At manual reduction without resection an intraluminal mass may either be too small to be palpated or masked by local edema of the bowel wall (83).

II.6.4.3. Bacterial and parasitic infection

There are many case reports of intussusception associated with bacterial infection. Tuberculosis has been mentioned, mainly in old series (164, 239, 249) or in reports of tropical intussusception (8, 49, 170). Salmonella (49, 164, 312) Shigella (107) and Yersinia (35, 358) were found occasionally. Reports of intussusception and Trichuris infestation (312) are rare. Tropical intussusception is often found in combination with Ascaris infection (8, 49, 170, 312, 357) and in combination with amebiasis (170, 269, 357) but most of the time in a minority of cases and found in an area where these infestations are endemic. Worms can form a bolus, Ascaris can attach itself firmly by its stoma to the intestinal wall and Entamoe-

ba can give severe inflammatory reactions in the bowel wall. Yet their role in the etiology of, even tropical, intussusception seems limited. It is striking that – although the importance of infection in the pathogenesis of intussusception has been emphasized so often – to my knowledge there has never over the years been an extensive controlled study of the relevance of bacterial and parasitic infection in intussusception.

II.6.4.4. Inflammatory bowel disease

Intussusception associated with acute colitis, ulcerative colitis and regional enteritis has been reported and is very rare. In contradistinction, intussusception in adult coeliac disease is not uncommon, the chance of finding it being related to the intensity of searching for it at small bowel barium examination. Most of these intussusceptions are of the enteric type, nonobstructive, short and transient (48, 295). There may be a relation to the characteristic small bowel dilatation in coeliac disease. On the other hand these intussusceptions may explain the idiopathic small bowel ulcer formation in coeliac disease. Most cases of intussusception in coeliac disease are asymptomatic. Complicated cases with obstruction and perforation have been reported.

II.6.5. Miscellaneous factors

II.6.5.1. Intestinal intubation

Intussusception is an infrequent but serious complication following intubation of the small intestine with a long indwelling tube (111, 250, 282, 314, 322, 346). Most of these intussusceptions are of the enteric type, more often jejuno-jejunal. They present most of the time as uncomprehended intestinal obstructions within the first two postoperative weeks. In the majority of cases tubes with an air-, water- or mercury-containing bag, like a Cantor (250, 314) or Miller-Abbott (322) tube, were involved. However intussusception with a Baker tube, lacking a bag at its distal end, has also been reported (282). One case of intussusception associated with intubation in a child has been described (346).
Most intussusceptions are antegrade and occur after withdrawal of the tube or while the tube is in place (111). The mechanisms of intussusception postulated are several. The bag acts as a stimulus for intestinal peristalsis and causes a spasm. The bag may also be gripped by peristalsis and serve as a leading point. Alternatively, as folds of small intestine become telescoped over the long tube due to ongoing propulsive peristalsis gripping an anchored tube, they may

become fixed by fibrinous adhesions. These fixed plications project into the lumen and may subsequently act as a leading point of intussusception (Figure 15). This can occur over the in situ tube or after its withdrawal or removal. Some intussusceptions associated with intestinal intubation are retrograde and cause symptoms after the tube has been removed. In most reported cases surgical treatment, often with resection, has been necessary.

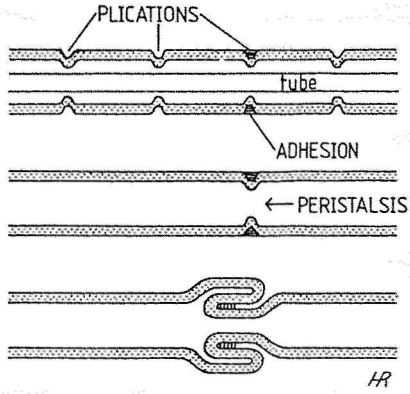

Fig. 15. Intestinal intubation causing intussusception through plication and adhesion formation.

II.6.5.2. Trauma

Reports on post-traumatic intussusception are rare. In former times Leichtenstern noted 8% of his series of intussusception due to trauma. Falor reviewed the literature and added one case of his own. He concluded that more factors than the trauma itself must have been involved. A recently ingested meal or severe fright could have played a role besides a violent central and adrenergic stimulation resulting in autonomic nerve imbalance and spasm (93). More recent reports are also rare. The reported time between the trauma and the intussusception sometimes runs to 10 months (!) (71, 164). Weilbaecher reported 5 posttraumatic intussusceptions in a series of 160 adult cases (366). Nagorney found 1 case in a series of 48 (228). Blanc described an enteric intussusception in an haemophiliac boy due to a hematoma from blunt trauma. Mourot and Holgersen both reported one case in a child after laparotomy for abdominal trauma (224, 144). Duncan e.a. reported 6 cases of intussusception as the cause of obstruction after laparotomy for trauma. These cases concern two factors. A post-traumatic situation and a post-operative situation. The clinical picture was similar to that of adult intussusception or post-operative intussusception. He

pointed out intestinal hematomas, abnormal peristalsis and suture lines, as predisposing factors although he found no such things in his cases. His data on intestinal intubation are insufficient to assess their importance (71).

II.6.5.3. Postoperative intussusception

Intussusception complicating the recovery period after different types of operations is a rare event in children and even more in adults. Two definitions have been used. Some include every intussusception, that has occurred after an operation (58, 60, 211, 218). Others exclude any intussusception that has presented itself after surgery for a previous intussusception, even at another location. Some also exclude any intussusception seen after surgery outside the abdomen (31, 80, 368).
Intussusception accounts for 5-10% of postoperative obstructions in children (58, 218, 267). It occurs after 0.08% of all laparotomies (80). Usually postoperative intussusceptions account for < 5% of all intussusceptions in children (80, 133, 328). West reporting the largest series until now (36 cases) calculated that 30% of all intussusceptions were postoperative (368). Cox calculated 16% (58). Guttman calculated 12% (125).
The following data are from the 5 largest series (58, 80, 211, 218, 368) and from literature reviews (60, 125). The patients reported, were aged 1 week to 18 years and the mean age varied between 17 and 30 months. About 70% is seen in children up to 2 years of age (60). Both sexes are equally affected (60, 218). These intussusceptions are for the major part (90%) of the enteric type. Ileoileal intussusceptions are twice as frequent as jejunojejunal.
Postoperative intussusception is noted in more than half of all cases following abdominal procedures of a diverse variety. Retroperitoneal operations, especially urologic operations (70, 328), account for 15% of cases and abdominoperineal procedures also account for about 15%. Following successful reduction of an idiopathic intussusception, 10% of all postoperative intussusceptions occurred. In about 5% intussusception complicated non-abdominal operations like cervical lymph node biopsy, revision of a ventriculo-atrial shunt and correction of coarctation of the aorta.
The time between the first operation and the laparotomy for intussusception averaged 8 to 11 days with a range from 1 to 26 days (58, 125, 211, 368). The length of time patients were symptomatic before undergoing re-operation, averaged 6 days (2-21 days) (211). Unlike postoperative bowel obstruction due to adhesion formation (the number one cause) that in 75% occurs more than 2 weeks after surgery (218), postoperative intussusception is encountered

within 1 week after surgery in 50-60% of all cases and within 2 weeks in 80-90% (80, 218, 368).
The symptomatology resembles postoperative obstruction due to adhesion. The symptom complex consists of abdominal distension (2/3 of cases) and vomiting (70%). Increased or persisting voluminous nasogastric drainage of bilious material is an important feature. Abdominal pain is not a prominent symptom, lacking periodicity and colicky violence, variable in nature and often difficult to interpret in the light of the recent prior operative procedure (211, 218). Rectal bleeding occurs in 10-15% (in contrast with the majority of cases with idiopathic intussusception). An abdominal mass is rarely palpated (<5%) (60, 211, 218). This must be attributed to the smaller bulk of the enteric intussusception, guarding and spasm evoked by palpation of a recently operated patient, abdominal distension and interference with palpation by wound dressings, drains and stoma's (211).
The diagnosis of postoperative intussusception can be made only if the index of suspicion is high. Abdominal radiographs are usually consistent with high-grade small bowel obstruction revealing dilated loops with multiple air-fluid levels. The most common opinion in the literature is that contrast studies of the colon are usually unrewarding in arriving at the diagnosis of postoperative intussusception since the lesion is usually located in the small bowel (58, 211, 368). Contrast studies of the upper gastrointestinal tract routinely fail to be of help in reaching the diagnosis pre-operatively. The passing of contrast material through the intussusception without delineating it has been reported (328). The use of ultrasound to demonstrate the intussusception may be rewarding but can easily be disturbed by intestinal gas (31). A small minority of postoperative intussusceptions can be treated by hydrostatic reduction.
At re-operation, most intussusceptions can be manually reduced (90-95%). Postoperative intussusceptions may be multiple so examination of the entire length of small bowel is necessary. Leading points are uncommon (about 10%, as in most populations) (60, 218, 368) and implicate inverted appendiceal stump, (anastomotic) suture line, polyp, lymphoid hyperplasia and traumatised intestinal wall (60, 80, 211, 218, 368). Much has been speculated as to the origin of most cases of postoperative intussusception. It may be related to disordered peristalsis induced by handling and drying of the bowel wall during operation, by adhesion, by anesthetic agents, postoperative administered drugs, neurogenic factors etc. Other factors, including concomitant chemotherapy and radiation, extensive retroperitoneal dissection, abnormal serum electrolyte levels and local hypoxia, have been implicated (60, 80, 211, 218, 368). The etiological importance of the use of long indwelling intestinal tubes

in many of these children (58, 211, 144) has not been emphasized enough.
Postoperative intussusception in adults is so rare that no percentage can be found in large series of postoperative small-bowel obstruction. On the other hand according to some authors it accounts for one third of adult intussusception (3, 302). There are several resemblances but also some striking differences between adult and childhood postoperative intussusception. In adults these intussusceptions are also mainly of the enteric type but jejunojejunal intussusceptions outnumber ileoileal several times. Intermittent cramping pain is a prominent clinical feature in adults. In the majority of cases resection is required (302). These adult intussusceptions are not related to intestinal tumour. Most of the time they are associated with intestinal intubation, adhesions and intestinal suture lines (7, 302). Cases after resection for Crohn's disease have been reported (137).

II.6.5.4. Intussusception after jejuno-ileal bypass

Intussusception of the by-passed segment after jejuno-ileal bypass for morbid obesity has been reported to be an infrequent complication. It occurs in 0.5 to 3.5% of the cases (172). Recognition of this complication led to the early admonition that mesenteric fixation of the closed end was essential to prevent later intussusception. Metal clips placed in the proximity of the closed jejunal end and adjacent mesentery have been advocated because radiographic separation of the clips would make intussusception more probable (325). Accumulation of intestinal fluids and gas proximal to the obstruction is absent in intussusception of the by-passed jejuno-ileal segment. Nausea and vomiting may occur but are probably reflex in nature. The most important symptom is crampy abdominal pain. It may start years after the bypass procedure and be recurrent in character. The presence of an abdominal mass and a high index of suspicion is necessary to make the diagnosis promptly. The combination with pregnancy forms a diagnostic problem and has been described by Labaille (361). The presence of residual obesity and emotional factors make the evaluation of the acute abdomen difficult and have resulted in erroneous initial diagnosis and significant delay in recognition of the problem (215). Ultrasonography and CT for routine evaluation of patients who had intestinal bypass operation, have proved to be reliable (172, 197).

II.6.5.5. Intussusception after gastro-jejunostomy

The incidence of jejuno-gastric intussusception has been estimated 0.015% of gastro-enterostomies (207). However, gastroscopic evaluation of 506 cases of gastrectomies revealed 8 chronic intussusceptions, otherwise misjudged (15). Most of these intussusceptions (75%) occur after gastro-jejunostomy without resection (33, 241, 356). They have been reported after a Billroth II type of gastrectomy, with or without a Braun anastomosis (241). Intussusception after Billroth I type gastrectomy and after Roux-en-Y anastomosis is very rare (15, 356). Gastric intussusception into the duodenum, caused by benign and malignant tumors (229) and mucosal intussusception into the duodenum have been reported but gastric intussusception through the stoma into the jejunum has not been described.

Anatomically, three types of jejuno-gastric intussusception are described (Figure 16): Type 1, afferent limb intussusception (antegrade) (10% of all cases); Type 2, efferent limb intussusception (retrograde) (80% of all cases); Type 3, combined afferent and efferent limb intussusception (10%). Type 3 occurs more frequently in the early postoperative period.

The onset of symptoms may be as early as three days postoperatively and as late as 35 years (15, 207). Clinically, the process may be acute or chronic recurrent. The clinical features of the acute type are epigastric pain of sudden onset, vomiting of food, then of bile, and then of blood, and also a palpable abdominal mass. Most acute intussusceptions are of type 2. The chronic form is characterized by recurrent abdominal pain relieved by intermittent vomiting. The alleviation of symptoms occurs with the spontaneous reduction of the intussusception. Probably the chronic form is never diagnosed, in many patients. Upper gastrointestinal series must be performed during the symptomatic attack. The differential diagnosis includes peptic ulcer, pancreatitis, anastomotic leakage or stricture, internal herniation, adhesions and tumour (208, 241, 356).

A plain film of the abdomen may show a scarcity of gas throughout the gastrointestinal tract and a homogeneous gas-outlined mass in the upper abdomen (356). Upper gastro-intestinal study shows a mass originating from the stoma with parallel curvilinear lines (207, 356). The acute form of jejuno-gastric intussusception must be treated surgically. A delay of more than 48 hours can increase the mortality from 10 to 50% (33, 34, 208). Chronically recurrent intussusception may be treated conservatively (34) although surgery is sometimes necessary.

The etiology of these intussusceptions is not well understood. This complication seems unrelated to the type of gastro-enterostomy or

size of the stoma and no one seems to be able to suggest any technical manoeuvre to prevent intussusception after gastric surgery. Many causal factors have been proposed: hyperacidity, long afferent limb, jejunal spasm with abnormal motility, increased mobility of efferent limb, vomiting, adhesions, dilatation of the upper jejunum, retrograde peristalsis, pregnancy (361) and other causes of increased intra-abdominal pressure (208, 241, 356).

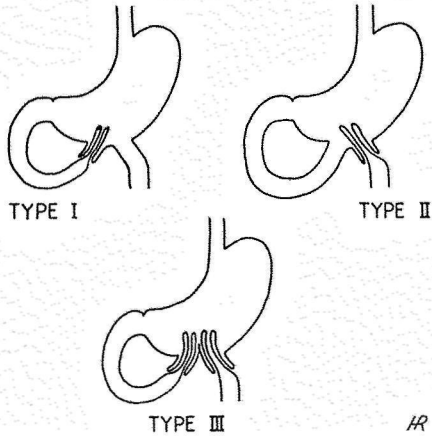

Fig. 16. Three anatomic types of jejunogastric intussusception.

II.6.5.6. Intussusception in chronic renal failure

The occurrence of intussusception resulting from intramural bowel haemorrhage is well documented in patients with Henoch-Schönlein's purpura, leukemia and in patients receiving anticoagulant therapy. Three cases of adult intussusception associated with chronic renal failure have been reported. These were believed to be a sequel of intramural bowel haemorrhage resulting from the coagulopathy related to platelet dysfunction in uremia, whereby an intramural haemorrhage formed the nidus for the development of intussusception (40, 379). No other anatomic lesion was found but the authors were unable to establish absolutely whether the haemorrhage was the cause, or the result of the intussusception. On the other hand intussusception in adults without demonstrable cause is rare and the association with chronic renal failure a striking coincidence.

II.6.5.7. Intussusception in pregnancy

With adhesive bands accounting for some 60 percent of intestinal obstruction seen in pregnancy, volvulus and intussusception follow as the second and third etiological considerations. Intussusception has been felt to account for 4-6% of intestinal obstruction in pregnant women (28, 361). However only 25 reported examples of this complication are encountered in the literature. The principal symptoms of intussusception occurring in pregnancy have been colicky upper abdominal pain, vomiting and constipation. These symptoms are noticed sufficiently often in otherwise normal pregnancy to delay recognition and treatment. Current hesitancy to order abdominal radiographs during pregnancy and the physiologic leukocytosis of pregnancy may also cause delay in diagnosis (28, 361). Rectal bleeding and an abdominal mass are infrequent symptoms.
Plain abdominal radiograph and barium studies are impracticable antenatally because of the radiation risk to the fetus. Ultrasonographic examination is the diagnostic tool of choice and may provide an early clue to the correct diagnosis. Misinterpretation of ultrasonography resulted in delay in two of three cases reported by Watson and Bourque (28, 361). Intussusception in pregnancy must be treated surgically. In ancient times it was associated with a high maternal and fetal mortality. Nowadays maternal survival is about 90% and fetal survival 60%.
Intussusception in pregnancy has been attributed to increased intra-abdominal pressure and to intestinal spasm caused by pressure of the uterus on the bowel. However a precipitating factor (Meckel's diverticulum, polyp, haemangioma, lymphoid hyperplasia, duplication cyst, suture line) has been found in 80% (361).

II.7. SYMPTOMATOLOGY

Introduction – General part

The clinical picture of intussusception varies greatly, depending on the course of the intussusception. These variations from one case to another are to be explained by a series of transitional forms between severe strangulation with total intestinal obstruction, an acute onset and rapidly progressing circulation disturbances, on the one hand, and slight strangulation with a chronic course, partial bowel occlusion and but little circulation disturbances or none at all, on the other. Any moment a chronic intussusception may turn into an acute case by further progress of the invagination or by obstructing bowel contents. However, most intussusceptions are

acute and the duration of the invagination determines the clinical picture, the pathologic changes in the bowel and the outcome of treatment. Thus, it is important to avoid any delay in diagnosis. Thorough knowledge of the value and meaning of the various symptoms is of utmost importance for the medicus practicus. A high index of suspicion and early recognition of the clinical picture are essential.
Since long ago, four cardinal symptoms have been characteristics of intussusception: abdominal pain, vomiting, rectal blood loss and an abdominal mass. Apart from these four symptoms intussusception is attended by several more or less specific accessory symptoms: pallor, drowsiness, fever, pulse rate elevation, abnormal peristalsis, free fluid, meteorism, abdominal resistance, facies abdominalis, Dance's sign, tenesmus, Wahl's sign, constipation, diarrhea, anal prolapse, abdominal distension, prostration, leucocytosis, dehydration and slackness of the sphincter ani.

II.7.1. Cardinal symptoms and signs

The onset of symptoms is usually sudden and in children the parents can tell the exact time at which the child cried out in extreme pain, turned pale, sweated and vomited. Such a paroxysm lasts for a minute or two and is followed by a period of relief. The attack recurs at frequent intervals of from ten to twenty minutes, sometimes longer. Between attacks the child may seem normal but also flaccid or prostrated. The duration of the attacks and of the free intervals varies considerably. At the beginning the attacks are usually long while the intervals are short. As the disease progresses the situation is reversed. Some children are not completely free of pain during the intervals between the attacks.
The chief complaint at presentation is vomiting in well over 40% of cases, followed by the passage of blood per rectum in also more than 40%. In only one fourth of cases abdominal pain was (one of) the chief complaint(s). Prolapse, distension, constipation, diarrhea, fever, prostration are variously given as chief complaints in over 10% of cases. Combinations were encountered (283). On the other hand evidence of abdominal pain announces the onset of intussusception a little more frequently than vomiting does. The appearance of blood per rectum is the first symptom in about 11% (281).

II.7.1.1. Abdominal pain

The pain in intussusception is usually characteristically intermittent, cramplike and at first very severe. Children with intussusception often scream loudly, fall in, writhe and turn about and draw

up their knees to their abdomen. Pain may appear to decrease in intensity late in the course of intussusception when intestinal dilatation (onset of gangrene) produces atony (83, 100, 147). In colocolic intussusception the onset of pain may be more stealthy (239). Colocolic intussusceptions are more often painless (164) than small intestinal intussusception. Small intestinal intussusception often gives rise to very moderate pain (due to relative width of the colon). Nyborg believed the attacks of pain in chronic intussusception to be extremely intense and longer, due to hypertrophic intestinal musculature exerting a strong pulling effect on the mesentery (239). Pain almost never lacks in chronic intussusception according to De Jong (164).
Colicky pain at some point in the course of the disease is almost invariably recognized in children over 2 and somewhat less commonly in infants. This presumably reflects equally on the articulateness of the children and the acuteness of the parents. In acute adult intussusception pain never lacks (164).
Sometimes the pain is continuous and not episodic, particularly in the peracute cases. Usually the pain is localized in the right abdomen or around the navel corresponding to the localization of the radix mesenterii. Most of the time the pain does not radiate.
Pain is a rather constant symptom. In 18 of 21 large series concerning various intussusception in children colicky pain was observed in over 75% of all cases (1, 10, 17, 32, 44, 62, 75, 110, 122, 133, 134, 156, 174, 186, 196, 201, 203, 220, 234, 256, 291, 323, 332, 336, 337, 362, 375).
Colocolic intussusceptions are more often painless (164). Ein found pain to be absent in 13% of his patients (78). In these cases the duration of symptoms before admission was twice as long, the hydrostatic reduction rate was 6 times as low and the bowel resection rate was 3 times as high as in the cases with pain. According to De Jong the mortality rate in painless intussusception is doubled but he contradicts Ein in respect of the duration of symptoms. De Jong never observed the combination of hyperperistalsis and the absence of pain. He assumes a bowel atonia on the basis of shock to be responsible (164).

II.7.1.2. Vomiting and anorexia

Vomiting is an initial symptom. The early vomiting is reflex, produced by a pulling effect in the mesentery appearing in connection with the paroxysm of pain. Early in the course of the disease the frequency and severity of vomiting is not related to the place of intussusception. Vomiting late in the course of the disease is less frequent in colocolic intussusception due to its relative width, a

competent ileocecal valve and its distal location (164). In the chronic form of intussusception, particularly in adults, vomiting is much less frequent (164). Vomiting is always a symptom in small intestinal intussusception, chronic and acute. The vomiting of intestinal obstruction is a late sign and should never be seen in a case of intussusception that has been properly handled. Faecal vomiting happens but exceptionally and, invariably, constitutes a bad prognostic sign (239).
Also vomiting is a constant symptom. In 30 of 32 large series concerning various intussusceptions in children vomiting was observed in over 70% of all cases (16 x >85%). (10, 17, 44, 62, 75, 105, 139, 140, 156, 196, 201, 203, 256, 270, 291, 315, 323, 332, 336, 362). Almost all infants vomit in the course of the illness and more than 80% of the older children vomit. In the infants vomiting tends to begin earlier in the course (164, 179, 281).

II.7.1.3. Rectal blood loss

The first examination of the bowels soon after the onset of symptoms usually shows normal faeces that is present distal to the intussusception. A small interval follows during which neither faeces nor flatus are evacuated. Usually the mesenterial incarceration will cause the formation of pathological intestinal contents consisting of serous fluid, mucus or blood. Many regard the discharge of bloody bowel contents as almost pathognomonic of intussusception in children. It is not only an important symptom for the physician but also often an alarming sign for the parents, causing the child to be brought to a doctor.
Characteristically blood is passed admixed with mucus in the classical currant jelly stool. At times, the stools consist almost entirely of mucus with only a slight admixture of blood. In other cases only guaiac positive stools are found. Sometimes there is copious passage of a thin bloody fluid (prune juice stool) or actually what appears to be clotted blood. The amount of blood lost externally is rarely great.
The space of time elapsing from the beginning of the disease to the time when blood is discharged is considered to be dependent on the rapidity with which the circulation disturbances progress, the length of the passage it has to run in the intestinal canal and the speed with which this is done. A bloody rectal discharge may appear within the first 2 or 3 hours or may not appear for a day or more. Ravitch found in 37% of cases bloody discharge in the first three hours (281). De Jong 63% within the first 12 hours (164). Thus rectal blood loss does not seem to be a late symptom. According to Nyborg the onset of rectal bleeding is evenly distributed in different

groups of duration of symptoms (239). Early discharge of blood was relatively rare. Bruce found a similar figure, only 16% had detectable bleeding within the first 12 hours (32).
In infants the intussusception is more often accompanied by bleeding (164, 239, 281, 315). Possibly this fact is also dependent on the fragility of the infantile intestinal wall and vessels or a quicker passage through the colon but a more frequent strangulation must be the main reason.
Colocolic intussusceptions are almost always accompanied by rectal blood loss. In small intestinal intussusception bleeding occurs in about half the cases. Reasons can be a longer passage, or bowel atonia. The time of admission can play a role. In adult cases and in chronic intussusception rectal blood loss is not common.
Ravitch found rectal blood loss in 95% of the infants and in 65% of the older children giving a 91% frequency in all cases (281). Comparable figures are only found in earlier reports (1, 122, 186, 249). Most authors report rectal blood loss in 50 to 70% (10, 17, 44, 62, 75, 105, 133, 134, 139, 140, 174, 196, 203, 220, 234, 270, 315, 319, 323, 332, 336, 337, 341, 362, 375). According to several authors rectal blood loss is not a consisting finding at all, reporting less than 40% of cases (9, 32, 110, 291). It is not always clear if these figures account for rectal blood as a symptom or also as a finding at physical examination. In about 10% the first evidence of blood is seen when the examining finger is withdrawn from the rectum, emphasizing the necessity for rectal examination.

II.7.1.4. Tumour and prolapse

The presence of an abdominal mass is a significant diagnostic finding in intussusception. At first the abdomen is flat or scaphoid and in this condition the mass may be visible. The abdomen is soft and nontender, although palpation of the mass itself may elicit a little tenderness and muscular resistance. The mass is tubular or sausage-shaped with its concavity towards the umbilicus determined by the dragging effect of the mesentery. Almost always there is an increase in size and firmness during a paroxysm of pain or during (bimanual) palpation. This should make palpation easier if it was not accompanied by spasm of the abdominal wall. Therefore palpation under general anesthesia usually is successful. The size and mobility of the mass varies according to its position in the intestinal canal. The mass may lie anywhere along the anatomical course of the colon but may be found almost anywhere in the abdomen. The palpation of a mass may be difficult due to its passage into the hepatic flexure, behind the liver and right costal margin, due to its localization in the pelvic region or due to ab-

dominal distension and spasm. Generally, it is well delimitable and without soreness. The discovery rate of the presence of tumour is low in small intestinal intussusception and in adult intussusception (164). The finding of an abdominal mass depends on the scrutiny of the examiner and therefore varies greatly. In seven reports of 38 large series an incidence of more than 80% was calculated (17, 122, 134, 186, 273, 315, 337) but in two other reports this percentage was well under 30 (9, 110). Most authors reported a tumour in 60 to 80% of all cases (25x >60%) (1, 10, 62, 75, 133, 174, 196, 201, 203, 249, 270, 315, 319, 332, 336, 375). The commonest site of the tumour is the right abdomen (almost 60%), particularly the right hypochondrium. In one fourth of cases the mass is left-sided and in about 15% epigastric (10, 17, 291, 332). In earlier reports a predominance of left abdominal masses was found related to a longer duration of symptoms (164). In accordance with this finding is a high proportion of cases in earlier reports in which the mass could be felt in the rectum (>20%) (1, 122, 187, 271, 284). In the majority of recent reports this figure is below 10% (32, 62, 75, 156, 196, 234, 291, 323, 336). Again, this emphasizes the necessity for bimanual examination. The rectal mass resembles the portio uteri.
In a small proportion of cases (<5%) the intussusceptum can also prolapse through the anus, attaining a length of up to 20-30 cm (32, 105, 122, 156, 234, 273). It is usually of a deep purplish colour and may be gangrenous. It has been mistaken for prolapsus ani, polyp or even haemorrhoids. It can be distinguished from prolapse of the rectum by the long separation between the protruding intestine and the rectal wall reaching up from 8 to 10 cm. According to De Jong prolapse is not a bad prognostic sign (164).

II.7.2. Accessory symptoms and signs

II.7.2.1. General condition: lethargy, shock, fever, dehydration, pallor

In the acute form of intussusception the general condition may deteriorate within a few hours. But the disease will rarely take such a course. During the attacks of pain paleness, uneasiness, cold sweat as well as small and rapid pulse indicate shock. But usually, at the beginning of the disease, the child seems perfectly normal between the paroxysms. Gradually a deterioration of the general condition will be increasingly noticeable. The complexion becomes ashen grey, the look absent, the eyes hollow and the mind blunted, the facies abdominalis develops. Many terms have been used for the accompanying mental state: apathy, lethargy, drowsiness, prostration, listlessness. This state is a characteristic sign of intussuscep-

tion, pointed out as early as by Hirschsprung. De Jong found this apathy ("as caused by autointoxication") to be an early symptom without prognostic significance (164). Some degree of this state is noticed in up to 50% of cases (134, 164, 272). The most pronounced form, usually described as shock although not precisely defined, is reported to be present in about 10% of all children (1, 10, 133, 134, 174, 196, 234, 336).
At the beginning of the disease the body temperature is often normal. This explains the differences in numbers for fever in the literature. With a cutoff point between 37.5 and 38.0 C the percentage of children with fever varies from 13 to 42% (10, 17, 164, 203, 256, 270, 281, 323, 337). Fever is both commoner and higher in infants. After successful operative or hydrostatic reduction high fever for 1 or 2 days is common. Outside the attacks of pain there is no significant rise in pulse rate in patients without fever (164). Dehydration is dependent on the extent of the vomiting and of the intestinal obstruction. The numbers given in the literature depend on the interpretation of the examiners. Some authors report a percentage of about 15% (1, 10, 17, 234, 323, 336). According to others it is about 45% (196, 270, 281). Ravitch emphasized the grave prognostic significance of dehydration (281). Initially paleness is present during the attacks, later also between attacks. Paleness is caused by a sympatic reflex. It is noted in about one fourth of all cases (10, 17, 164).

II.7.2.2. Constipation and diarrhoea

In most instances of intussusception, once the child has evacuated the bowel content distal to the intussusceptum, feces and flatus are not passed and intestinal obstruction is complete. Constipation is noted in less than 10% of cases in series with a low mean duration of symptoms (110, 174, 234). With increasing mean duration of symptoms the number of children with observed constipation rises to about 30% (270, 281, 336).
What is apt to be more confusing is the occurrence of diarrhoea after the onset of intussusception. This creates the danger of a misdiagnosis of dysentery. In large series diarrhoea is noticed in 5 to 15% of cases with few exceptions (10, 32, 110, 156, 174, 201, 234, 270, 281, 323, 332, 375). Beasley and Suita reported 31 and 26% respectively (17, 336).

II.7.2.3. Anorexia and weight loss

The feeding history of the children after the onset of symptoms is of interest. About 50% of children take food but less than 10% are able to retain it (139, 271).
Hirschsprung described the classic case of intussusception as arising in the well nourished infant and is quoted as never having seen this disease in a malnourished child (32). However, as in our own series, a recent report of a large prospective study suggested that a significant number of children with intussusception were in fact undernourished (159, 160). Bruce could not confirm either concept (32). At birth children with intussusception are of average weight (202).

II.7.2.4. Upper respiratory tract infection and otitis media

In the light of etiological considerations it is noteworthy that about one quarter of all children with intussusception have an upper respiratory tract infection or otitis media at admission (10, 32, 108, 261, 270, 273). Gierup reported only 5% while Ross reported over 50% respiratory infections (110, 292, 293).

II.7.2.5. Signs of bowel obstruction

Visible peristalsis as a symptom of mechanical ileus may, of course, be seen in intussusception but it is not very common (273, 164). This symptom has been observed more frequently in chronic than in acute intussusception, probably due to considerable hypertrophy of the intestinal musculature (164, 239). The intussusception rarely forms a protuberance which is visible. The abdomen, early in the disease, is flat or actually scaphoid. However, abdominal distension may appear at any stage of the disease as a sign and a consequence of complete intestinal occlusion. It may obscure the intussusception mass. In the majority of large series distension is found in about 25% of children with intussusception (10, 17, 156, 234, 239, 256, 273). Radiological evidence of intestinal obstruction on plain abdominal radiograph has also been found in about 25% (110, 291).
Increased bowel sounds can be heard in about 20% of cases (164, 256). Pollet noted the absence of bowel sounds in 12% (256).

II.7.2.6. Miscellaneous symptoms and signs: Peritonitis, Dance's sign, Wahl's sign, anal slackness

Typically, the abdomen is soft and nontender although the child may wince when the intussusception is palpated and in such instances a little muscular resistance over the mass may be felt. Tenderness is noted in about one third of all cases (32, 270, 273). Fortunately signs of generalized peritonitis are found in a small minority, less than 5% (17, 110, 164).
A flattening of the right fossa iliaca which felt empty when palpated, was emphasized by Dance as a symptom of an ileocecal intussusception. Most of the time the empty space will be occupied by distended small intestinal loops. Usually this sign is found in less than 10% of all cases (32, 164, 239) but Swenson reported it in one-fourth of his patients (337).
The finding of a distended bowel loop proximal to the intussusception tumour is called Wahls's sign. One finds two tumours, one firm-elastic and periodically increasing in firmness, another soft and balloon-like. Wahl's sign is rarely reported (164). In former times great diagnostic significance was attributed to slackness of the anal sphincter but nowadays not much attention is paid to this symptom (239).

II.8. DIAGNOSTICS

In many cases the diagnosis of intussusception can be reached early on the basis of a thoroughly taken history and an accurate physical examination. However, early manifestations of intussusception are frequently deceptive and some patients have an atypical presentation. A safe diagnosis on clinical grounds alone may be difficult or impossible. Even in cases with fully developed intussusception symptoms and signs may remain vague or atypical for a considerable period of time. In these cases a high index of suspicion is mandatory to make the diagnosis. Complementary examination may be adopted if the presence of an intussusception is not certain or even not deemed likely. More than laboratory tests, imaging studies have proved to be of much help in the detection and confirmation of intussusception.

II.8.1. Laboratory tests

Laboratory aids such as haemoglobin, guaiac testing for occult blood, white blood count and differential are usually of no great diagnostic benefit (281, 362).

Anemia is a rare complication in children (<5%) (174) but in adults with chronic intussusception it may be present. Guaiac testing for occult blood has its diagnostic significance in chronic intussusception too (259). In Raudkivi's study the leucocyte count averaged 15.9 x 10.9/l with a range from 6.5 to 44.9 x 10.9/l (270). A high percentage of children has a leucocytosis (174, 259, 273, 337). Ponka found the higher leucocyte counts in infants in whom intussusception had been present for some time (259). Ravitch calculated that a white blood count of 12 x 10.9/l or higher was as frequent in patients who died as in survivors and he concluded that leucocytosis was of no prognostic significance (273). About 10% of children has a white blood count over 20 x 10.9/l (44, 174, 259, 273). According to Ching this finding and a shift to the left is associated with a high incidence of gangrenous bowel (44).

II.8.2. Imaging studies

II.8.2.1. Plain abdominal radiograph

Plain abdominal radiographs are used to separate cases of suspected intussusception, suitable for hydrostatic reduction, from those demanding surgical treatment. In the presence of free, intraperitoneal air and pronounced intestinal obstruction, barium enema is often precluded. However, plain radiographic diagnosis of intussusception is controversial. The following signs have been described to be associated with intussusception:

- occurrence of gas-fluid levels in normal calibre or dilated intestinal segments.
- decreased gas and fecal content in the right colon
- presence of a discernible soft-tissue mass at the site of the intussusception
- presence of a gas-outlined apex
- gas-filled loops of the small intestine at the usual site of the right colon.
- a cone-shaped, gas-filled terminal loop of the ileum or colonic loop (88, 110, 165, 303, 369).

With the first 5 criteria a diagnostic specificity of 94%, sensitivity of 75% and accuracy of 87% can be reached (27). Others doubt the diagnostic significance (193).

Plain abdominal radiographs of patients with intussusception show gas-fluid levels in about 60% of cases (1, 88, 90, 139, 149, 196, 30). The combination of gas-fluid levels and dilated bowel loops is seen in about 30% (17, 106, 110, 174, 196, 236, 259, 263, 291, 323, 369). Strikingly small amounts of gas and fecal material in the right colon characterized three fourths of all cases (87, 88, 110).

A soft tissue mass is observed on the survey film of about 55% of all patients (87, 110, 196, 236, 323, 369) and a gas-outlined apex in only 15% (110). Gas-filled small intestinal loops at the usual site of the cecum or ascending colon are visible in half the cases (88, 110). Although very helpful, none of the signs is specific for intussusception, and several authors consider plain abdominal radiograph inconsistent and unreliable for diagnosis. Yet, Ekloff (90) stated that a positive diagnosis of an intussusception is possible in most of the suspected cases. To achieve this he safely recommended a policy consisting of two or three radiographs on wide indication in all patients with abdominal complaints. The increased dose of radiation and cost of examination more than counterbalanced the radiation of previously liberally performed barium enemas and the cost of the associated time of hospitalization. Nevertheless there always remains a certain number of cases in whom the safe exclusion of this disorder calls for a confirmatory barium enema.

II.8.2.2. Contrast enema examination

Although often the clinical history and physical findings are sufficiently typical for diagnosis, barium enema usually is used to confirm intussusception. Other contrast material may also be used like air or water soluble contrast. This method is said to have a high sensitivity and specificity (306). However, as far as I know, no study of the accuracy of barium enema has been reported, not even in times in which in many clinics every child with intussusception was operated on and every hydrostatic reduction was confirmed by laparotomy. Some intussusceptions reduce very quickly as the barium enters the colon at low pressure and without attempt at hydrostatic reduction being made, leaving no time to make a radiograph and leaving the radiologist in doubt as to whether he/she saw an intussusception or some phantom. The presence of fecal material can also interfere.
There is a little discussion concerning the contraindications for contrast enema examination. Peritonitis, the combination of high fever and radiographically overt small bowel obstruction and severe constitutional symptoms and signs are absolute contraindications. However, until the seventies, in several clinics, especially in the United Kingdom every child was operated on the more clinical suspicion of intussusception (62, 256, 332, 336). Barium enema was omitted to avoid any risk of perforation and delay in surgical therapy. In many countries during this century, gradually, barium enema examination was used to confirm the suspected clinical diagnosis of intussusception in every child. Ekloff was able to exclude about 25% of children on the basis of plain abdominal

radiography findings. In his study supplementary barium enema was performed a) in patients under 6 months of age, b) in patients presenting with scant intestinal gas and in particular if the caecum could not be outlined, c) in patients with a possible mass lesion of the colon, d) in cases with suggested obstruction of the gut presumably due to intussusception and e) in cases of discrepancy between the radiographic observation and the clinical manifestations (90).
Barium enema examination is especially fit for diagnosis of intussusceptions with a large colonic component. The film may show one of three configurations (Fig.17). Owing to edema of the apex or contraction of the receiving layer the contrast pillar may assume an abrupt terminal shape, the so-called amputation configuration. A concave ending of the barium (a cap formation) may also point to complete colonic obstruction. These two pictures may show a notch in their centre. A filling defect in the head of the barium which frequently presents as a central defect, surrounded by two parallel contrast streaks, connected by closely placed curled linear spiral pattern, is called the coil spring appearance. These transverse, narrow stripes of contrast may also be less close. They represent a streaky puddling of the barium in the compressed lumen between the apex and the receiving layer (38, 303). All these pictures may show barium entering the lumen of the entering layer. This central canal is usually narrow and has longitudinal rugae (239, 303).

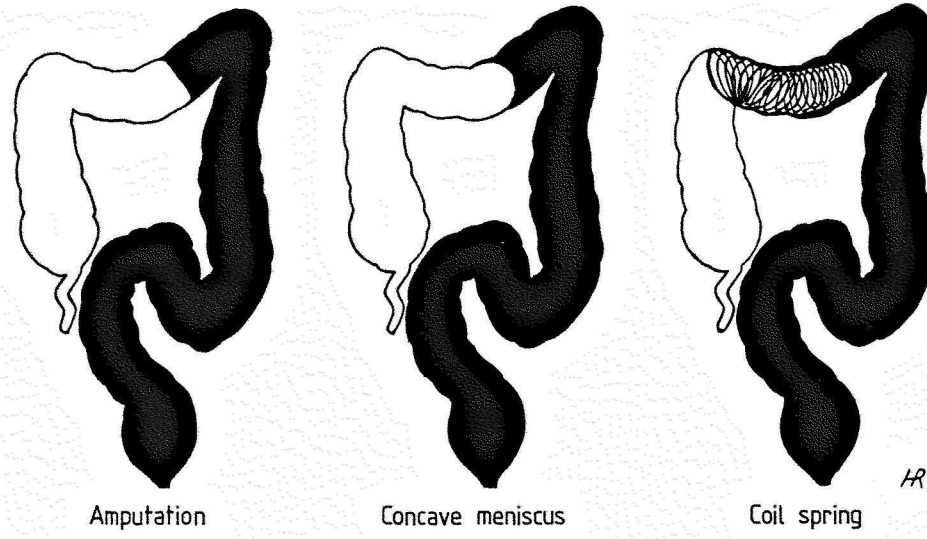

Fig. 17. Barium enema configurations of intussusception.

When the contrast penetrates far down in the caecum, a peculiar drop of the flexures and a shortening of the colon may be observed.

If there is a leading point, it can often be demonstrated at the apex of the intussusception (303).
In recent series the apex configuration by barium enema examination has been encountered in the terminal ileum in less than 5%, in the caecum or colon ascendens in about 35%, in the colon transversum in about 45%, in the colon descendens in about 10% and in the colon rectosigmoideum in less than 10% (110, 216, 236, 270).
The differential diagnosis with an obstructing tumour or polyp is usually no problem as the configuration shifts toward the ileocecal valve under the pressure of the enema. On the other hand, the differential diagnosis of a small irreducible, nonobstructive cecocolic, ileocecal or appendicocecal intussusception is very comprehensive and may be difficult. The following diagnoses have been taken into account in the past: Bolus of ascaris lumbricoides (171), huge blood clots (117), malignant tumour (carcinoma, carcinoid, lymphoma, metastasis), benign tumour (mucocele lipoma, adenomatous polyp, leiomyoma, villous adenoma, neurofibroma), inflammatory tumour (ulcerative colitis, regional enteritis, solitary cecal ulcer), infectious tumour (abscess, ameboma, tuberculosis, diverticulitis), inverted appendiceal stump, submucosal haemorrhage and endometrioma (109, 303).
Barium enema is not pre-eminently suited to disclose small intestinal lesions. In particularly favorable cases the contrast may pass through the ileocecal valve, divulging the apex of a pure small intestinal intussusception or after hydrostatic reduction the ileoileal component of an ileo-ileocecal/colic intussusception. The difficulties in distinguishing between ileocecal, ileocolic and colocolic intussusception in the colon ascendens have been emphasized. A topical diagnosis is unascertainable in these cases. It is impossible to arrive at any conclusion with regard to type from the length of the disease history and the distance covered by the intussusceptions (239, 303).

II.8.2.3. Small bowel follow-through meal

Small bowel follow-through meal is particularly apt to diagnose small bowel intussusception. Contrast material introduced orad to an isoperistaltic intussusception may enter the lumen of the intussusception or not. In the latter case the typical features of obstruction are demonstrated. The proximal gut is dilated and the sudden change in caliber of the gut marking the beginning of the intussusception is easily demonstrated, sometimes showing the shape of a bird's beak.
If the central canal and the intussuscipiens fill, the apex will often be demonstrated as in barium enema examination (153, 238, 30).

However, sometimes the patency of the central canal may lead to the misdiagnosis of a normal image. Small bowel follow-through meal is not used routinely. Most reports on its successful use concern small intestinal intussusceptions and longstanding ileocolic intussusceptions with atypical symptoms.

II.8.2.4. Computed tomography

Computed tomography has shown to be useful in atypical cases of intussusception. In adults with chronic abdominal complaints of pain (59, 251, 316) especially if an abdominal mass was found (66 157, 197) intussusceptions with a colonic component were demonstrated by CT. Other, rare cases like postoperative intussusception (335), intussusception of the excluded loop after intestinal bypass for morbid obesity (197), chronically recurrent intussusception in a child (192) and small intestinal, adult intussusception have been diagnosed by CT (212).
Already in early reports a complete and accurate description of the characteristic features of the CT image of intussusception was given (66). These features include:
- a central, eccentric soft tissue mass, representing the intussusceptum.
- A crescentic fat density representing the mesentery of the involved bowel.
- An outer rim with bandlike areas of high density intermingled with somewhat curvilinear regions of low density (layering or stratification) representing the returning and the receiving layer.
- Spicules of contrast material peripheral to the intussusceptum correspond to the coil spring sign in barium enema examination.
- A leading mass may be seen or the apex may be delineated by contrast material or by air (59, 66, 157).

These images were also demonstrated in experimentally induced intussusceptions in dogs (59, 157).
CT cannot be advocated as a primary tool in the diagnosis of intussusception but the use of CT is an appropriate measure in the patient with a history of partial small bowel obstruction and non-diagnostic standard contrast medium examination. In the presence of an abdominal mass, scans obtained at 1 cm intervals at the level of the mass, may be more conclusive.

II.8.2.5. Other imaging techniques

Reports on the use of endoscopy (154, 209) and angiography (43, 189, 311) for the diagnosis of intussusception are sporadic. The use of radionuclide imaging of intussusception is nonspecific and has

been mainly limited to experimental studies in dogs and mice (72, 183, 243). The use of ultrasonography will be discussed extensively in Chapter III.8.

II.9. DIFFERENTIAL DIAGNOSIS

In most textbooks is emphasized that the diagnosis of intussusception presents little difficulty provided the physician has the condition in mind, particularly in acute cases (84, 100, 147). There are, however, cases that differ in various ways from the usual picture (22, 94). Atypical presentation often results in delayed diagnosis and increased morbidity (281). A high index of suspicion in regard to intussusception and a sense of urgency in the presence of the possibility of intussusception, should reduce the number of diagnostic errors. Particularly the absence of one of the four cardinal symptoms, abdominal pain, vomiting, rectal bleeding and abdominal mass, may cause a significant diagnostic problem (164). In the absence of abdominal pain in intussusception the duration of symptoms before admission is twice as long, the hydrostatic reduction rate is 6 times as low and the bowel resection rate is 3 times as high as in cases with pain. Too much stress may be laid on failure to feel a mass by abdominal palpation or by rectal examination. The mass may be misinterpreted as an enlarged liver (274). The mass can also disappear as in recurrent intussusception (192) or enlarge improbably rapidly (349).

Other errors in diagnosis are based on a number of misconceptions including failure to recognize the typical history or to perform rectal examination, prolonged or recurrent history with the wellbeing of the child, atypical age, presence of an underlying disorder, bright rectal bleeding or bloody diarrhea considered to be of infectious origin and incomplete bowel obstruction. The issue may also be clouded because the patient was already in the hospital with another disease (22, 266, 281).

The symptoms of intussusception overlap those of many other conditions (371), particularly in chronic or chronically recurrent intussusception, small intestinal intussusception and intussusception in older children and adults. When pain and vomiting constitute the only features of the symptom picture, the intussusception may be mistaken for commonplace gastro-enteritis, acute nutritional disturbance, dyspepsia or abdominal pains connected with throat infections (22, 118, 134, 164, 239, 274, 276). Bloody bowel movements and abdominal cramps accompanying colitis may usually be differentiated from intussusception because the pain is less severe and less regular and because the infant is recognizably

ill between pains from the time of onset (118, 134, 164, 239, 274). From the point of view of differential diagnosis Henoch-Schönlein purpura may occasion insurmountable difficulties. It contains abdominal pains, vomiting and intestinal bleeding in its symptom picture. In a differential diagnostic sense, cutaneous bleeding and haemarthros when occurring, are important signs. It is important to keep in mind that intussusception may be a complication of this disease. Contrast enema and ultrasonography may be of diagnostic help but a hematoma in the intestinal wall may produce an impression of an intussusception tumour (22, 118, 134, 164, 239).

Bacillary dysentery may also give rise to problems of differential diagnosis as against the acute intussusception. Besides abdominal pain and vomiting, malodorous, frequent and bloody diarrhoea accompanies dysentery. This diarrhoea contains faecal matter and bile-coloured substances, a fact which renders them distinguishable from the entirely bloody or slimy bloody intestinal evacuations in the case of intussusception (164, 239, 274, 276).

Appendicitis can cause any bewilderment from the point of view of differential diagnosis, especially in older children. Sometimes, in the absence of bloody stools and a mass, sufficient spasm and hypersensitivity to pressure in the right fossa iliaca is present to simulate an acute abdominal condition like appendicitis. Such soreness, however, rarely accompanies intussusception. On the other hand, Dance's sign and bloody stools are absent in appendicitis (22, 164, 239, 274, 276).

One of the more common errors is in making the diagnosis of constipation. It will often be accompanied by severe pain of a pronouncedly intermittent character. Accumulation of faecal matter may be mistaken for an intussusception tumour. Vomiting and blood discharge are mostly absent (22, 164, 239, 274). Meckel's diverticulum may contain abdominal pain with occasional vomiting and rectal blood loss in its symptom picture, simulating intussusception (164, 274).

A malignant bowel tumour, bleeding cavernoma, a bundle of helminths, a volvulus and an incarcerated hernia may show all four cardinal symptoms of intussusception (164, 239). Several causes of small intestinal obstruction, polyps, abdominal tuberculosis and actinomycosis may also resemble intussusception (164, 239, 274). Other diseases, mentioned in the differential diagnosis or mistaken for intussusception are cyst, sickle-cell crisis, otitis media, plumbism, colon duplication, enlarged liver, rectum prolapse, meningitis, hydrops of the gall-bladder, ovarian cyst, congenital intestinal stenosis, lowered kidney, renal tumour, peptic ulcer, appendiceal abscess, type 1 familial hyperlipoproteinemia and even sigmoiditis

with hermaphrodosia spuria feminia with first menses (164, 239, 274, 276).

II.10. TREATMENT OF INTUSSUSCEPTION

As early as the times of Hippocrates and Praxagoras a controversy may have existed between the advocates of conservative treatment on the one side and those of surgical therapy on the other side. Until the mid-nineteenth century the disease was almost universally fatal but occasionally responded to conservative measurements such as inflation of the bowel. The sporadic cases, treated surgically ended fatally almost without exception. Not in the least because most patients were presented for surgery in a late stage of the disease. In the second half of the 19th century things changed. Progress in asepsis, anaesthetics and surgical technique made the mortality rates of surgical treatment go down. On the other hand Hirschsprung published the first of a series of reports dealing with the systemic reduction of intussusception by hydrostatic saline pressure. Although his first report dates back to 1876, his results, far more superior to those achieved by primary operative treatment, were not accepted until 1905 (141, 142). Around 1927 several reports on the successful use of hydrostatic reduction appeared in Australia (Hipsley), Scandinavia (Olsson), France (Pouliquen) and the USA (Retan, Stephens) (281). In the first half of the 20th century a lively discussion developed between advocates and opponents of conservative treatment especially as the mortality rates of surgical treatment gradually decreased to less than 1%. Thanks to Ravitch, hydrostatic reduction became the primary treatment of choice in the USA in the 50's and 60's (271, 272, 273, 274, 275, 276, 277, 278, 279, 280, 281). In spite of the efforts of Zachary conservative treatment did not gain a place in many clinics in the UK (382, 383, 384). Even in 1985 a discussion concerning the primary treatment of intussusception could be followed in the Lancet (188).
Nowadays the standard primary treatment of intussusception in most clinics over the world is conservative. Surgical treatment is reserved for hydrostatically irreducible cases, adult intussusceptions, perforated intussusception most chronic cases and many recurrent intussusceptions.

II.10.1. Operative treatment

Once the decision to apply surgical treatment is taken, laparotomy should be done as soon as possible. However, formerly, stress was laid on the necessity for spectacular haste in moving such patients

to the operating room. It is now generally accepted that the immediate need is for intravenous fluid administration to restore fluid balance. Gastric suction and proper coverage to reduce loss of body heat should be taken care of. On the other hand, there is a direct relationship between the results of attempts at manual reduction and the duration of symptoms. So, it is wise to operate as early as possible.

The localization, direction and size of the incision depends on the localization and extend of the intussusception. A high Mc Burney or transverse right midabdominal incision is recommended for intussusception around the ileocecal valve not extending beyond the midtransverse colon (9, 32, 112). For larger intussusceptions this kind of incision offers insufficient operation space (239, 337). The alternative is a more generous midline incision (122, 239, 281). The incision should permit insertion of the operators index and third finger into the abdomen. The first part of the reduction should, as far as possible, be conducted within the abdominal cavity. The method of manual reduction is named after Hutchison. The intestine is grasped, being compressed, gently but firmly, over the apex, just distal to the intussusception. In this way the intussusceptum is milked back by progressive compression in the direction of the collar of the sheath (taxis). The remaining mass can then be pulled outside the abdomen where it is handled with greater facility. The reduction is continued by taxis rather than an attempt to disengage the intussusception by traction. If the bowel reduces easily and rapidly there is no need to deliver the intussusception or to question the viability of the bowel. Sometimes the strangulation at the collar, owing to edema, will constitute a very troublesome obstacle in the way of reduction.

If the bowel stubbornly resists reduction it is preferable to resect it at once rather than to use excessive force and risk rupturing the bowel and contaminating the wound and peritoneal cavity. Attempts to stretch the neck of the intussusception with instruments result in disaster and incision of the neck of the intussusception invites contamination of the operative field. It is rare that an intussusception, so difficult to reduce that it requires these strenuous methods, will prove not to require resection because of gangrene or doubtful viability. The reduced bowel will frequently be of a beefy, red or blue-black colour and yet, after several minutes of observation, it will be seen to have a good tone, improved colour and to be capable of peristalsis. Bowel of questionable viability can often be improved by covering with a saline pack.

The indication for bowel resection is threefold:
- the intussusception is irreducible
- the bowel is perforated or nonviable or has a doubtful viability
- the presence of an organic lesion that needs resection.

A variety of methods has been proposed from primary resection and anastomosis to staged operations of various kinds with exteriorization, extra-abdominal amputation, double barrelled ileocolostomy or shortcircuiting anastomosis (122, 164, 239, 281). Nowadays results are vastly superior to those obtained in the first half of the century. An adequate current experience is available with direct resection and primary anastomosis with recovery rates as high as with the less attractive staged procedure and this more straightforward and definitive procedure seems to be established as the procedure most frequently advocated and performed (239, 281).

In early as well as recent reports concerning series from children hospitals where more than 88% of all patients with intussusception was operated on, the resection percentages were around 10 to 15% of all cases (1, 9, 20, 44, 62, 112, 139, 140, 168, 185, 187, 203, 249, 256, 259, 270, 315, 323, 332, 336). This resection rate is the same in series with hydrostatic reduction rates of about 50% (11, 17, 23, 32, 57, 68, 75, 105, 129, 149174, 234, 252, 291, 323, 362, 375). Very low resection rates are reached in series characterized by early admission, aggressive diagnostic measurements and a small proportion of cases (about 30%) coming to surgery (24, 110, 184, 196, 201). High resection rates (22-44%) are, not rarely, mentioned, even in recent reports (10, 44, 259, 263, 341).

An appendectomy is indicated in all cases explored through a right lower quadrant incision to avoid future misunderstandings. Resection of a Meckel's diverticulum is obvious. Enterotomy is indicated with a suspected intraluminal leading point at palpation. During an operation an abnormal movability of the caecum will not rarely be observed. This has been considered to have a direct causative connection with the origin of the intussusception. In order to prevent recidivism, fixation of the caecum to the abdominal wall or limiting its movability by wrinkling of the mesentery have been proposed. Suturing the terminal ileum to the ascending colon has also been presented to be successful in preventing recurrence (36). These prophylactic operations have more and more been abandoned, entailing risks of intestinal incarceration on account of bridles and adhesions as they do (374). The frequency of recidivism is very low, even irrespective of these measures. Relapses occur even after fixation of the caecum (239).

Before primarily closing the abdomen it is advisable to ascertain whether the reposition is complete, the circulation disturbances are reversible and no probable causes of the intussusception are over-

looked. Special attention should be paid to the fact that intussusception at the ileocecal valve may be accompanied by a second intussusception more proximal in the small intestine. Proper examination of the whole ileum and jejunum is required (131).

II.10.2. Conservative treatment

II.10.2.1. Spontaneous reduction

Not all intussusceptions need treatment. An incidence of about 5% for spontaneous and complete reduction of intussusception between the time of diagnosis and the time of laparotomy in patients undergoing primary operation has been reported in most series giving numbers on this subject (1, 9, 62, 75, 106, 174, 185, 187, 336, 375). Higher percentages are given concerning spontaneous reductions, not proven by laparotomy (139, 140, 184). An edematous and cyanotic bowel at laparotomy in combination with a clear clinical picture is considered to be an almost 100% proof. Spontaneous reduction would be due to distension of the bowel loop situated close to and orally of the intussusception, to antiperistalsis in that loop, or to traction of the mesentery. The use of opioids or morphinomimetics for general anaesthesia may be important because of resulting relaxation of the intussuscipiens.

II.10.2.2. Sloughing of the intussusceptum

From the earliest days of our knowledge of intussusception it has been known that in some instances the intussusceptum becomes gangrenous and is sequestered, the bowel fusing at the neck of the intussusception and retaining its continuity. This is called sloughing, auto-amputation or sphacelation of the intussusceptum. The slough or sphacelus is passed by rectum, with recovery following. This requires the rapid production of gangrene and sequestration before the patient succumbs to toxemia and intestinal obstruction. The sloughing as a rule, does not take place before the fifth day (164, 239). Such instances, which today are rare and evidence of neglect, represented almost the only hope for survival in the 19th century. Most reported cases concern more chronic intussusceptions in adults. Close to 50 per cent developed (either) perforation with peritonitis or intestinal obstruction due to progressive cicatricial constriction (42, 239).

II.10.2.3. Hydrostatic reduction

There will be general agreement today that a barium enema study still is the conclusive diagnostic test when intussusception is suspected and that it should be undertaken in the expectation of applying it therapeutically if the diagnosis is confirmed. The method has been scrupulously described by Ravitch, based on clinical experience (1954) as well as animal experiments (1948) (272, 275). Many authors report to have followed this method in broad outline (10, 38, 67, 68, 75, 140, 184, 201, 309, 323, 362, 375).

As soon as the presumptive diagnosis of intussusception is made, the child is taken to the fluoroscopic room. At the same time the operating room is advised to prepare for operation. The participation of the surgeon who will do the operation, if one is required, may avoid unnecessary delay. A nasogastric tube must be in place and intravenous fluids must be running. Without anesthesia a Foley bag catheter is inserted into the rectum and the balloon is distended with 20 to 40 ml of air according to the size of the patient. If the intussusception presents outside the anus, it has to be replaced into the rectum digitally to permit the introduction of the catheter. It is essential that the catheter be ungreased, so that it may be less readily expelled. The catheter is pulled down against the levators. Traction on the catheter will at times be required to produce a tight seal. The buttocks and the thighs are strapped together with adhesive.

A large reservoir, obviating delays during the treatment, is filled with barium sulphate suspended in lukewarm, normal saline solution. With the canister no higher than 100 cm above the table the contrast solution is allowed to run into the bowel. As soon as the intussusception is encountered the barium flow is interrupted for a documenting film. At this point, the rounded head of the advancing barium column suddenly becomes concave, forming a meniscus around the point of the intussusception. As pressure increases, the meniscus lengthens, the horns extending until suddenly the intussusception is pushed back and the meniscus flattens out again. The process is repeated, sometimes with extreme rapidity, until the intussusception is reduced to the cecum and through the ileocecal valve. The fluoroscope need only be monitored intermittently to observe the progress of the reduction in stages, and not continuously. As long as there is any change at all in the head of the barium column or in the barium leaking ahead of it between the coats of the intussusceptum, the procedure should continue. Delays of about 10 minutes may occur at one point or another, especially the splenic and hepatic flexure and (most often) at the cecum. It is most important to maintain uninterrupted hydrostatic pressure with

smoothly flowing barium. As long as the barium column continues to advance or to change in outline or consistency or configuration, one can assume that progress, however stubbornly and slowly, is being made and the treatment should be pursued. If complete reduction is not achieved after a stubborn arrest of more than 10 minutes, the child is permitted to expel the enema and the effort is repeated. A number of three attempts with a total time of about 45 minutes is advocated. The best interest of the largest number of patients will be served by fixing a limit to the time that one continues the enema after a stubborn arrest.
No one – surgeon, radiologist, or pediatrician – lays a hand, however gentle, however educated, however lightly encased in a lead glove, upon the abdomen to manipulate the intussusception.
Complete reduction of an intussusception at the ileocecal valve is diagnosed by

1. The free flow of barium well into many loops of the small bowel.
2. Disappearance of the intussusception mass.
3. Clinical improvement of the child.
4. Passage of feces or flatus together with the expelled barium.

An abdominal survey film is always taken 15 minutes after the enema has been evacuated since, with the colon largely empty, the small bowel is more positively distinguished. Sometimes it is possible to observe that the barium passed 3 or 4 inches up into the ileum and than stopped while the characteristic meniscus is still discernible, the ileoileal component of a ileo-ileo(ceco)colic intussusception. According to some authors (217) this type of intussusception can be distinguished on initial encounter of the intussusceptum by barium running along side the apex. This has been called 'the dissection sign' (98) and has been associated with irreducibility. Others deny that this sign is typical for ileo-ileo(ceco)-colic intussusception (239) and that hydrostatic reduction attempt is deemed to fail (217).
The post-evacuation film in successfully reduced intussusceptions should reveal one of the following features:
1. Free reflux to the small bowel, increasing during attempts at evacuation. 2. Poor evacuation ability.
In irreducible intussusception one or more of the following findings are to be expected. 1. Propagation of the contrast medium in oral direction is arrested by the intussusception. 2. There is a constant filling defect in the contrast medium in the proximal colon or in the terminal ileum 3. There are persisting signs of small bowel obstruction. There are dilated, gas-containing small bowel loops above the suspected obstruction. 4. Strikingly complete evacuation (85).

One may be confronted with a typical defect in the lateral cecum, opposite the ileocecal valve, representing invagination of the anterior lateral aspect of the cecum including the lateral half of the anterior tinea coli. In these cases one is justified in assuming a cautious 'wait and see' attitude (63, 99, 255). On repeat enema at 24h this caecal filling defect has usually disappeared. However a persisting intraluminal medial filling defect may be a persisting intussusception, an edematous ileocecal valve, feces or an intramural or mucosal mass. Hydrostatic reduction may also be performed under ultrasound guidance (359, 365).

If for a period of 10 minutes there is absolutely no change in the shape, outline, position, or density of the barium, the procedure is discontinued and the child taken to the operating room (271, 275, 277, 279, 281). In a quarter of those operated on, the intussusception will be found to have been completely reduced or reduced to the right side of the colon or caecum where a small right lower quadrant incision and a minimal manipulation will be all that is required. Thomas reduced only to the caecum to operate all his patients (341).

The technique of hydrostatic reduction shows important differences depending on the cautiousness, thoroughness and persistance of the administrator. Like Hirschsprung in his times several authors do not shrink away from bimanual manipulation of the intussusception through the intact abdominal wall (110, 174, 216). From Ravitch's experimental work it appears safe to raise the pressure to not over 100 cm of water to avoid reduction of gangrenous bowel and perforation of the bowel (272). Nevertheless many authors exert a pressure of 150 cm of water (110, 149, 216, 236, 239). The number of attempts at hydrostatic reduction may even be 10 (216). The use of a 'second enema' after one to several hours for cases of incomplete reduction has been advocated (52, 196). The use of different contrast media and premedication is so much at debate that it will be discussed separately.

Differences in the technique of hydrostatic reduction, in experience and in selection of patient material are of important influence on the success rates reported in the literature. In most publications a success percentage between 50 and 70 or slightly less is mentioned (10, 11, 17, 24, 32, 57, 68, 69, 75, 105, 106, 129, 149, 156, 174, 234, 236, 253, 281, 291, 323, 337, 362, 383). A percentage of about 80 has been reported from clinics where the intussusception is manipulated through the abdominal wall (110, 216), where a delayed, second attempt is made (52, 196), where an extensive experience is present (110, 184, 196, 201, 352, 354), where most patients submitted to an attempt at hydrostatic reduction have a duration of symptoms of less than 24 hours (110, 184, 201, 216) or

where the barium column is raised over 150 cm (110, 216). Rather low percentages for successful attempts at hydrostatic reduction (< 40%) are reported from hospitals where the incidence of intussusception was low and the experience with hydrostatic reduction was limited (112, 270, 341), where hydrostatic reduction was not vigorously attempted (259, 375) where a limited availability of a skilled radiologist was reported (203, 341) or where a strong preference for surgical treatment existed (9, 44, 341).

The percentage of successful attempts at hydrostatic reduction has been reported to vary with several factors. The presence of roentgenologic signs of intestinal obstruction is associated with a 20% lower reduction rate than unselected material (110, 196, 236). According to Frye success is not related to ileus, according to Potts the reduction rate is only 8% in the presence of bowel obstruction (106, 263). The success rate of hydrostatic reduction has been related to the location of the apex of the intussusception. The numbers given by Gierup, Ein '71 and Nordshus suggest that the reduction rate is lower as the apex approaches the rectum (75, 110, 236).

There is also a relation between the success rate and the duration of symptoms. The reduction percentage of intussusceptions with a duration of symptoms of less than 24 hours is about 80% (32, 57, 110, 149) although a percentage near 60 is also reported (105, 236, 323). The second day the success rate is about 20% lower (32, 110, 149, 236). Intussusception with a duration of symptoms of more than 24 hours has a hydrostatic reduction rate of one third or less (10, 57, 105, 133, 149, 196, 323). An acceptable percentage is only reached in experienced hands (32, 110, 236).

The success percentage in children under 6 and under 12 months of age have been found to be lower than in older children (110, 236, 323).

Most authors reporting on the hydrostatic reduction of recurrent intussusception, found an even higher success rate than in primary intussusception (75, 105, 201, 236). It seems worthwhile to treat recurrences hydrostatically but the presence of a leading point must be excluded afterwards. In former years it has been stated that intussusception caused by a lead point cannot be reduced by hydrostatic enema and necessitates operation (77, 26). Recently, however, the successful hydrostatic reduction attempts of intussusceptions caused by an organic lesion have been described (82, 110). Intussusceptions of the ileo-ileo(ceco)-colic type are known to be hardly reducible (110263). The reported hydrostatic reduction rates for intussusception in the absence of abdominal pain are very low (78, 203, 323) with one exception (10). According to Gierup reduction rates are low in the presence of vomiting or rectal blood loss (110).

The criticisms of the conservative treatment which have been levelled from time to time, fall under the following headings:

1. The original diagnosis may be uncertain.
However, diagnosis by fluoroscopy is simple and accurate. In addition, pediatricians are much more likely to make a tentative diagnosis of intussusception when to do this, commits them merely to an enema rather than an operation. The result is earlier definitive diagnosis and treatment.

2. There may be uncertainly of reduction.
An erroneous diagnosis of complete reduction is rare if the criteria for complete reduction are strictly followed. If doubt exists about the reduction the abdomen must be opened. In many such cases operation may disclose the fact that reduction actually has been complete.

3. The rate of recurrence is higher.
The usual figure given for recurrence in intussusception following operation is around 3%. After conservative reduction it is said to be about 10%. If there is recurrence it is likely to be recognized easily and early and the child can again be submitted to a barium enema.

4. A causative lesion may be overlooked.
At operation leading points can usually be recognized and dealt with. A lesion of this sort might be missed with a hydrostatic reduction but most of these lesions can be found by careful examination of the postevacuation film. It is fortunate that most of these lead points are not such that, in themselves, cause serious concern. A serious exception, the lymphosarcoma at the ileocecal junction, is rare.

5. A dangerous delay may be caused.
There is a problem of time wasted by an exhausting procedure in patients who may have to go on to surgery after attempted hydrostatic reduction has failed. However many a 'critically ill' child has been treated successfully hydrostatically within a few minutes. On the other hand, time, needed to prepare the operating room and required to apply resuscitative measures, may at the same time also be used for an attempt at hydrostatic reduction without delaying surgical treatment.

6. There is a danger of bowel perforation.
The incidence of bowel perforation during hydrostatic reduction is less than 1%. Many of these perforations were encountered before

an attempt at reduction was made. There is clinical and experimental evidence that additional perforations are avoided by a correct technique.

7. Nonviable bowel may be reduced.
With a pressure of 100 cm of water one does not reduce gangrenous bowel. Irreducibility is determined by adhesions between the sheaths and by the degree of edema. Both factors increase with time and become more effective in preventing reduction as the damage to the bowel increases. It is important to note that the stress of the pressure is borne by the intussuscipiens which usually remains viable to the end.

8. Experience in hydrostatic reduction is needed.
It is true that results are better in hospitals where a high incidence of intussusceptions provides a great experience in hydrostatic reduction. However, inexperienced physicians may also reach a considerable success rate in a safe way.

9. Purely ileal intussusceptions can not be treated.
A high index of suspicion is needed to detect the enteric component of an ileo-ileocolic intussusception on the postevacuation film. In these cases sometimes reduction is possible. Purely enteric intussusceptions will not be submitted to hydrostatic reduction as they are usually not detected by barium enema. These children will be operated on because of intestinal obstruction.

10. Radiation exposure to the young individual is an disadvantage.
The roentgen exposure must be only fractional and intermittent using a small field. The facility of image intensification and timer must be available. The covering of genitals is difficult if not impossible.

The advantages of hydrostatic reduction are obvious.

1. Avoidance of an anaesthetic and of laparotomy.
The avoidance of an anaesthetic must be outweighed against the use of medication during hydrostatic reduction, advocated by many authors. After operative reduction of intussusception, there is a significant incidence (about 10%) of complications, woundabscess formation, wounddehiscence and late mechanical intestinal obstruction due to adhesion.

2. Early discharge from hospital.
If not performed in an out patient set up, hydrostatic reduction

allows discharge of the cured child from the hospital, within 24 hours, having escaped the operative risks and discomfort. This leads to the avoidance of the psychological effects of prolonged hospital admission.

3. Partial reduction.
Even if the intussusceptions should prove to be incompletely reduced, the more of the intussusception that is reduced by the enema, the less remains to be done in the operating room. The problem of reducing an intussusception found to be in the right colon or in the cecum is quite another from that of reducing the intussusception in the rectum or sigmoid entirely around the colon.

4. Lower resection rate.
It is of interest that the number of resections is lower in series treated primarily by barium enema. Presumably this indicates that a number of the resections that are otherwise performed are made necessary by operative trauma to the bowel during the reduction or represent resections of bowel mistakenly thought to be nonviable.

5. Normal alimentation is resumed within four hours.
Certain clinical contraindications to attempted enema reduction have been recognized such as a history of symptoms of more than 24 hours duration, the presence of signs of advanced intestinal obstruction and a bad general condition of the patient. These contraindications will be discussed in chapter III.2.

II.10.2.4. Pharmacological support of reduction

Controversy exists concerning the use of pharmacological agents to improve the rate of successful reduction. In accordance to Ravitch's original recommendations most authors hold the view that the use of drugs is unnecessary (281). However in some reports, to facilitate reduction the use of sedatives, analgesics, relaxants or general anaesthetics has been advocated.

Analgesia and sedation.

Zachary, using a combination of Atropine and a barbiturate, reached a success rate of 61% (383). Frye used meperidine (1mg/lb i.m.) and a barbiturate (1mg/lb i.m.) to reach 52% (106). Touloukian administered a combination of meperidine (1mg/kg), promethazine (1mg/kg) and chlorpromazine (0.5mg/kg) or a barbiturate. His success rate was 68% against a remarkably low 36% in his control group (347). Bruce reported a 65% success rate using a combina-

tion of meperidine, promethazine and chlorpromazine (32). These rates are not high compared to figures from clinics where no drugs were used. The most important advantage seems to be the possibility to use a correct reduction technique on a quiet patient.

General anaesthesia.

Minami suggested that under deep sedation or general anesthesia attempted reduction with manipulation through the abdominal wall may be possible. In this way he could report a 82% success rate (216). Smith, using Ketamine or an inhalational anaesthetic saw his success rate go from 40 to 82% (188). Collins improved his success rate from 68 to 84% by administering a second attempt at hydrostatic reduction under a general anesthetic (52). These figures are high but can also be reached without medication (110). The drugs decreasing smooth muscle activity in the bowel by decreasing vagal tone, by ganglionic blockade or by competing with acetylcholine on its smooth muscle receptor site, may have been of influence. But also a correct hydrostatic reduction technique on a baby that does not cry or struggle, may have been of influence. A second attempt as in Collins study may benefit from the time delay in which edema may diminish by improved venous drainage from a partially reduced intussusceptum.

Glucagon.

In the early 1970's glucagon became a popular means of inducing smooth muscle relaxation, considered an advantage in many radiologic procedures. At first there were anecdotal reports indicating that the administration of glucagon may facilitate hydrostatic reduction of intussusception. Using glucagon (0.05mg/kg i.m. or i.v.) in combination with morphine sulphate (0.2mg/kg i.m.) Hoy increased his success rate from 61% to 84% in an uncontrolled study (151). In contrast to this Haase in experimentally accomplished intussusception demonstrated faster reduction and earlier return of blood flow to the involved bowel but no rise in successful reduction. He used glucagon (0.05mg/kg i.v.) in dogs under barbiturate anesthesia (30mg/kg) (126). In a double-blind study Franken failed to confirm any benefits of glucagon (0.05mg/kg i.v.) (103). Mortensson, in a controlled (not double-blind) study did not find glucagon (0.05mg/kg i.m.) combined with morphine, to improve the rate of employed reductions nor the time necessary for reduction to take place. He found an indication for a limited benefit of adjuvant glucagon after two unsuccessful barium enemas before a third attempt is performed (222).

Miscellaneous.

Liu, after injection of a combination of diazepam (250-300ug/kg i.v.) and hyoscine butylbromide (400ug/kg i.v.), used a second attempt at hydrostatic reduction under general anesthesia. His success rate was 79% (196). Grahl changed from none to atropine-premedication and found the reduction rate to rise from 45% to 85% (116). Meuli recommends the use of glucagon (0.5mg/m^2 i.v.) in case peroperative manual reduction is not possible (214).

II.10.2.5. Gasreduction of intussusception

The use of gas to reduce intussusception has been known since the time of Hippocrates and possibly has been advocated through the centuries (21). As the use of barium reduction under fluoroscopic control gained acceptance, simple insufflation of air per rectum, controlled by manometric readings of the pressure and fluoroscopic observation has been proposed, but did not find imitation in the Western world (97). In contradistinction, in the Far East this method is widely practiced ever since. Very high reduction rates (>90%) have been reported, concerning huge series of patients, built up in the last 25 years (124, 377, 385). Since these reports a controversy exists centering on the use of barium or gas under pressure control and fluoroscopic guidance.

Technique of gasreduction.

An appropriate-sized (8-16 French) Foley catheter is placed in the rectum, the balloon inflated with 15-50 ml and the buttocks are securely taped. Air or oxygen is inflated with a handbulb or using an electrical airpump or an oxygen tank. The gas pressure is controlled by a sphygmomanometer, a U-tube mercury manometer or automatic pressure selector. Gas is inflated continuously or intermittently, to maintain the pressure at 60 to 80 mmHg, maximum pressure being up to 120 mmHg, Under fluoroscopic control a steady flow of gas is delivered to the colon. Once the intussusception is recognized progressive reduction is achieved under a constant pressure. Sudden flooding of multiple small bowel loops with air indicate reduction of the intussusception. At this moment auscultation at the site of the intussusception reveals a loud 'whooshing' noise. No progress in reduction requires more pressure but distension instead of progressive reduction demonstrates poor tolerance of the colon to such a pressure which should be reduced immediately (123, 124, 254, 339, 377, 385). The contraindications of gas reduction are essentially the same as for barium enema

reduction. Gas reduction has been combined with manipulation through the abdominal wall and is performed with sedation or under general anesthesia (124, 339, 377, 385).
In publications from China successful reduction rates of well over 90% have been reported. More recent reports from Western countries showed reduction rates similar to the best results reached with barium enema reduction (123, 124, 154, 339, 377, 38). The better results may be attributed to earlier diagnosis, less severely ill patients or the more sophisticated skills of the radiologic and surgical team (104).
The reduction time needed is 5 to 10 minutes and the fluoroscopy time is less than 1 minute. Perforation rates and mortality rates are well under 1%, similar or lower to barium enema reduction (123, 124, 254, 339, 377, 385). Gas reduction of intussusception is a simple, effective and safe method. Specific advantages are a very accurate pressure control and the avoidance of barium peritonitis in case of perforation. There are some disadvantages. Some doubt the diagnostic accuracy of fluoroscopy using air insufflation in relation to intussusception and to the presence of a lead point. A controlled study is needed to determine the most effective method in the treatment of intussusception, barium enema reduction or gas reduction.

II.11. COMPLICATIONS

II.11.1. Mortality

In the mid-nineteenth century intussusception was almost universally fatal but occasionally responded to inflation of the bowel with a bellows or to enemas. Progress in anesthesia, surgical technic and pre- and postoperative care prepared the way for the surgical treatment of intussusception. However, early in the 19th century, the mortality from surgical treatment was still approximately 90% (337). At the same time, Hirschsprung, using anesthesia, reduced the intussusception by salt water enema and had a 39% mortality rate (142). In the first half of the twentieth century the mortality rates reported by advocates of hydrostatic reduction (239, 281) were somewhat lower than those reported from clinics where primary surgical treatment was therapy of choice (122, 133, 138, 143, 186, 220, 249, 319, 332). In both kinds of clinics an important contribution to the mortality figures came from the group of children treated with bowel resection. In the sixties after further progress in technic and preoperative preparation the death rates were essentially the same in clinics with high (24, 129, 253, 355) and low (23, 174,

375) hydrostatic reduction rates and in clinics with primary surgical treatment (1, 20, 259, 337).
Since 1970 the reported mortality rates vary between 0 and 3% for primary surgical treatment (9, 62, 112, 168, 185, 203, 256, 270, 336) and for clinics with high hydrostatic reduction rates (10, 17, 68, 69, 106, 110, 149, 196, 234, 341, 362) and for those with low rates (57, 105, 156, 291, 323) with few exceptions (139, 184, 306). Today, there is essentially no mortality from intussusception treated in the best pediatric surgical clinics, except in children already irretrievably moribund at the time of admission or with complicating disease. A comparative study of current mortality statistics in series treated conservatively or surgically is therefore of little value.

II.11.2. Bowel perforation

Perforation of the bowel during attempt at hydrostatic reduction of intussusception is uncommon. The number of case reports is inversely proportional to its incidence. In reports of large series its occurrence is mentioned sporadically (14, 75, 110, 185, 195, 201, 205, 235, 291, 367). Its estimated incidence is under 1% (79, 152). Barium peritonitis resulting from bowel perforation during barium enema is a serious complication. Contamination of the peritoneal cavity with barium and bacteria causes an extreme peritoneal reaction and the formation of marked adhesions (173, 178, 288, 310, 386). It has been recommended that the enema be given with water soluble contrast material or gas rather than barium (6). However, if managed correctly, no mortality has to result (79, 115) as it has in earlier times (119, 128, 386). In 25 cases of bowel perforation during barium enema reduction of intussusception, reported in the last 25 years, no child died.

II.11.3. General complications

Complications following surgical treatment of intussusception may be divided according to the time interval and to the fact whether secondary laparotomy is required. Early complications requiring surgery are bowel perforation, wound dehiscence and evisceration, intestinal gangrene, anastomotic leakage and intestinal obstruction secondary to adhesions. These complications are reported in about 5% or less of all cases of intussusception treated surgically (1, 110, 112, 133, 156, 203, 234, 270, 291). In another 5% relaparotomy must be done because of intestinal obstruction due to adhesions after the immediate postoperative period (9, 17, 57, 75, 168, 196, 256, 273, 306). So, early and late complications, requiring laparotomy, occur in up to 10% of all cases of intussusception, treated

surgically (1, 9, 17, 75, 105, 110, 133, 168, 196, 203, 234, 256, 270, 273).
Early complications not treated by laparotomy include wound infection, pneumonia, phlebitis, otitis media, anuria, sepsis, convulsions, local peritonitis, prolonged postoperative ileus, enterocolitis and intractable diarrhoea (1, 17, 133, 174, 203, 270, 319, 337). They may account for up to 2% of all cases treated surgically (196, 203, 273, 306). Incisional hernia occurs in about 1% of all cases (133, 203, 319, 341).
Postoperative complications are more frequent in those cases where the duration of symptoms is longer than average (75). Complications occur more frequently in those patients in whom there have been diagnostic difficulties (17). There are also more complications in children whose intussusception require more than a manual reduction with or without an accompanying appendectomy (75).

II.11.4. Recurrent intussusception

Recurrent intussusception is a well recognized, although uncommon, sequel to successful reduction of an intussusception, whether it be by hydrostatic pressure or operative intervention. Reported recurrence rates range from 0 to 21% (16, 68, 76, 86, 136, 239, 259). with a combined average of about 4% (136, 321). After a recurrent intussusception, the risk of having a subsequent one is increased to about 20% (16, 76, 86). Most recurrent intussusceptions occur between a few hours and 3 years after successful reduction of the initial intussusceptions. Early recurrence is much more frequent after hydrostatic reduction. In one-third or more of cases, the recurrence occurs within 6 months of the initial hydrostatic reduction. Recurrent intussusceptions are diagnosed sooner than the first intussusception although there are fewer presenting signs and symptoms (especially rectal bleeding is lacking more frequently compared to first episodes) (16, 76, 86). The appearance of the abdomen at conventional radiography is essentially the same in initial and recurrent intussusception. The single important feature is complete evacuation of the bowel following the initial enema, observed in approximately one-third of the cases with early recurrence (76).
The recurrence rate after hydrostatic reduction of intussusception varies between about 5 and 10% (10, 75, 106, 129, 149, 196, 355, 362). However, much higher figures in large series have been reported (68, 201, 236, 323). When a recurrence is discovered within a very few hours of hydrostatically reducing a previous one, the likelihood of reduction having been incomplete must be considered. Although the opposite has been asserted (77, 281) the

successful hydrostatic reduction of an intussusception does not exclude the presence of a lead point (82). So recurrent intussusception after hydrostatic reduction may well be caused by a lead point overlooked at the time of the first episode. Indeed, a high incidence of leading points has been reported in series of recurrent intussusception, mainly treated surgically (20% Thorndike, 36% Beasley) (281, 16). The incidence of leading points in series treated mainly conservatively, is similar to that seen in series also treated operatively (76, 86).

The incidence of recurrent intussusception following surgical reduction of ileocolic intussusception is reported in the 0 to 5 % range (9, 10, 20, 62, 75, 133, 187, 319, 332, 336, 362). The adhesions created by the surgical manipulation of reduction of the intussusception and appendectomy may explain the reduced recurrence rate after surgical reduction of an intussusception. This advantage of surgical treatment is offset by the inevitability of some cases of mechanical intestinal obstruction due to adhesions. One should always bare in mind that recurrent intussusception may exist on the base of a suture line, a new polyp or a remnant of gastric tissue after resection of a Meckel's diverticulum. Recurrent intussusception after ileocolic resection for intussusception has never been reported. The major part (> 90%) of these recurrences do not occur in the immediate postoperative period (313, 321). The vast majority of recurrences following reduction of ileocolic intussusception is also ileocolic. In most cases, the etiology is not at all clear. Rarely an enteric intussusception, part of multiple intussusceptions and overlooked at the first laparotomy, presents as an early recurrence. This indicates the need for thorough inspection of the whole small intestine after manual reduction of an intussusception (131).

The treatment of recurrent intussusception is still subject to debate. The propagators of surgical treatment point to the high number of leadpoints, especially in older children. The more so as this lead point may well be a lymphosarcoma (81, 363). The advantages of hydrostatic reduction are obvious and have been pointed out before. The success rates of hydrostatic reduction of recurrent intussusception have been reported to be higher than of their original series (> 70%) (16, 76, 86). In children under 2 years of age with recurrence following a previously successful barium reduction, a further hydrostatic reduction should be attempted. Where a previous operation has failed to demonstrate a lead point, recurrence can be safely treated with a barium enema. In children over 2 years of age with recurrence, hydrostatic reduction of recurrence may also be attempted on the condition that it is accompanied by an exact examination of the reduction and evacuation films and that it is followed

by a thorough search for a lead point using small bowel follow-through investigation.

Indications for surgical treatment in recurrent intussusception include: 1. More than one recurrence where no prior operation has been performed. 2. Pathological lead point known or suspected from previous operative findings or clinical features of Peutz-Jeghers syndrome. 3. More than one recurrence after surgical treatment for intussusception (to avoid repeated hydrostatic reduction eventually resulting in laparotomy, often with resection). Various techniques for preventing recurrence have been utilized (36). Attempts to fix the ileum to the ascending colon, the cecum or the appendix to the abdominal wall have met with only fair success. After these procedures recurrences as well as bowel obstruction due to internal herniation have been reported (136, 374). If necessary ileocecal resection provides a definite solution for recurrent intussusception without further recurrences.

Chronically recurrent intussusception may form a distinct clinical entity. Ravitch has stated that about 7% of his patients had had an entirely similar attack at intervals of 10 days to 6 months before the attack that brought them to the hospital (281). The typical clinical picture of chronically recurrent intussusceptions is characterized by bouts of cramping abdominal pain (30-40 minutes duration) during a period of 1 month to several years, alternating with painfree intervals. Other characteristics are occult gastrointestinal bleeding and transient bowel obstruction. Later, an acute condition necessitating an operation often results. These intussusceptions seem to occur more frequently in older children and adults. They are of the enteric type, in many instances multiple, and extending over a short distance. They may present in different bowel coils. Small bowel follow through examination may reveal these intussusceptions but is often negative. At laparotomy the vitality of the bowel is intact and a lead point is not infrequently found (176, 340, 373).

Transient small bowel intussusception has been described in a minimum of 20% of adults with coeliac disease, routinely examined by a specific bowel meal (48). However, the incidence in the normal population of small bowel intussusception with quick spontaneous reduction not coming to medical examination is not known and may be significant.

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CHAPTER III

Own observations and investigations

III.1

Atypical Characteristics of a Group of Children with Intussusception

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Abstract. Reijnen, J.A.M., Festen, C., Joosten, H.J.M., and Wieringen, P.M.V. van. (Department of Pediatric Surgery, University Hospital, Nijmegen, The Netherlands). Atypical characteristics of a group of children with intussusception. Acta Paediatr Scand 00.

We describe the characteristics of a group of 140 children with intussusception. They differed strikingly from the classical picture of intussusception given in textbooks and in publications concerning large series. We found a low incidence of intussusception, especially in infants and young children. There were far more older children, the delay was significantly longer and there was a very high percentage of leading points. These factors explained our low hydrostatic reduction rate. The weight of most children was under the fiftieth percentile. The percentage of small bowel intussusceptions was somewhat higher. These differences are important in the light of early diagnosis and evaluating the results of treatment. Keyword : Intussusception.

Introduction

According to the literature intussusception has a classical clinical picture. Typically a child, more often a boy, less than two years of age who has previously been well nourished and in good health, suffers a sudden abdominal pain. Vomiting may occur either simultaneously with the pain or soon there after. Within 24 hours mucus or blood, or both may be passed together with thin bowel movements. Usually the child is presented within 24 hours from the onset of symptoms. In most cases an abdominal mass can be palpated. As a rule intussusceptions occur in the region of the ileocoecal valve. In a small proportion of cases it is possible to demonstrate a local abnormality leading the intussuscepted gut at its apex (1-12). We report about the characteristics of the group of children treated for intussusception in our two hospitals. These characteristics differed strikingly from the above described classical picture.

Materials and methods

The records of the pediatrician and/or (pediatric) surgeon of all 140 patients under the age of 15 years, admitted to the St.Radboud Hospital and to the St.Canisius-Wilhelmina Hospital in Nijmegen from 1968 to 1988 in whom the clinical diagnosis of intussuscep-

tion was supported either by radiological evidence or by laparotomy, were subject to a retrospective study. The data listed in Table 1 were compared with those from textbooks and well documented publications concerning large series from important pediatric centres with an upper age limit from 12 to 18 years, totaling more than 2800 cases (1-12).

Results

In Table 1 the data from our hospitals are compared with data from the literature. There were 93 males and 47 females. Their ages ranged from 1 day to over 14 years. The mean age was 3 years. The age distribution of our series is shown in figure 1.
With the exclusion of nine cases of chronic intussusception the children were presented with a mean delay of 47 hours. The mean duration of symptoms in children over 2 years of age was almost 3 days. Colicky abdominal pain (119 cases), vomiting (111 cases) and an abdominal mass (94 cases) were the most important clinical features. Bloody stools were lost before admission or found at physical examination in 48 cases. In 38 cases diarrhoea was reported. The weight of the child was under the tenth percentile in 46 cases and under the fiftieth percentile in 85 cases. All chronic intussusceptions were in children over 3 years of age. Six chronic intussusceptions were of the enteric type. Recurrent acute intussusception could not be ruled out with certainty in one case. Infrequent daily attacks of abdominal pain (8 of 9 cases), sporadical vomiting (7/9), marked weight loss (8/9) and a high incidence of leading points (6/9) characterized chronic intussusception.
48 Children were successfully treated by hydrostatic reduction, a successrate of 48%. 92 children were operated on. In 13 cases at laparotomy the intussusception was found to have reduced spontaneously. Of sixteen intussusceptions, entirely located in the small bowel, four were in children under 2 years of age. A predisposing factor was present in 12 of all enteric cases.
Bowel resection because of irreducibility or gangrene was performed in 15 cases. Five Meckels diverticula were resected. Enterotomy was carried out nine times for biopsy or excision of an anatomic abnormality.
In 34 cases a predisposing factor precipitating the intussusception was found (Table 2). The incidence under 2 years of age was 13% and over 2 years of age 39%. 31 leading points were found by palpation and enterotomy or at histological examination of the resected bowel.

Table 1. Group characteristics of 140 children with intussusception

l.b. = life births

Characteristic	Present data	Data from literature	References
Incidence	1.1/1000 l.b.	0.9 – >4/1000 l.b.	2-4
Gender ♂ : ♀	2 : 1	2 : 1 or 3 : 2	1-10
Weight	61% < 50th%	well-nourished	3,5,7,10
Age	56% < 2 yr	74% < 2 yr (63 - 87%)	3-10
Delay	51% < 48 hr	81% < 48 hr (64 - 90%)	7-10
Colicky pain	85%	85% (76 - 94%)	3-10
Vomiting	79%	81% (63 - 91%)	3-10
Bloody stools	34%	52% (16 - 67%)	3-10
Diarrhoea	27%	20% (6 - 31%)	3-10
Abdominal mass	67%	64% (24 - 87%)	3-10
Type of intussusception	86% at the ileocoecal valve	91% at the ileocoecal valve (88 - 98%)	3-7
Leading point	22%	8% (6 - 10%)	3-10
Resection	11%	14% (4 - 18%)	3-10

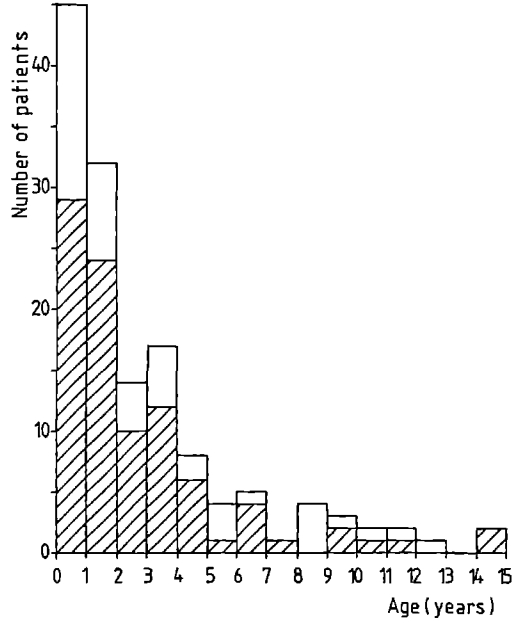

Fig. 1. Age distribution of 140 children with intussusception.

The presence of a leading point was suspected or proven pre-operatively in 2 cases of Peutz-Jeghers syndrome, 2 cases of Henoch-Schonlein's purpura, 1 case of an intussuscepted appendix stump and in 1 case of mucoviscidosis. In the latter case an ulcer of the coecum was found at the apex. In the other case of mucoviscoidosis inspissated, hard stool in the coecum extending into the terminal ileum precipitated the intussusception. In older children after hydrostatic reduction small bowel follow through examination and/or barium enema showed no additional leading point. There was a predisposing situation in 3 cases. In the case of severe nephrotic syndrome intramural hematoma formation precipitated intussusception. One post-operative intussusception occurred 1 day after appendectomy and the other 3 days after correction of a hernia hiatus oesophagi.
There was no mortality. During hydrostatic reduction no complications occurred. Anastomotic breakdown (1 case), wound dehiscence and evisceration (2 cases) and adhesive small bowel obstruction (6 cases) warranted reoperation after surgical treatment. There were 6 recurrences after successful hydrostatic reduction and one recurrence after surgical treatment. It was possible to reduce the intussusception by repeat enema in three cases.

Table 2. Predisposing factors in 140 cases of intussusception

Predisposing factor	Number of cases
Meckels diverticulum	8
Polypoid lymphoid hyperplasia	8
Appendix (stump)	3
M. Peutz-Jeghers	2
Mucoviscidosis	2
M. Henoch-Schönlein	2
Duplication cyst	2
Hamartoma	2
Fibroid polyp	1
Atresia	1
Post-operative phase	2
Nephrotic syndrome	1

Discussion

The incidence of intussusception in the Netherlands is low, about 1.3 per 1000 life births. We calculated an even lower incidence of intussusception in our region. Especially intussusception in children under two year of age seems to be less frequent than has been described previously (3-10). As a result, children in our district present relatively more often with intussusception at an older age.

Our high percentage of older children may have contributed to the long mean delay in diagnosis and treatment. The more so as the clinical picture in older children is often atypical and conducive to delay. Rectal bleeding is uncommon in older children and an important percentage exhibit only one or two of the four principal features of intussusception (11,13). The proportion of chronic intussusception does not differ from figures in the literature (13,14). Another delaying factor may have been the small proportion of cases with rectal blood loss. This symptom is often alarming and leads to the correct diagnosis (3,10). On the contrary, considering the mean delay of 47 hours one would expect a higher proportion of cases with rectal blood loss. An explanation could be a higher incidence of non-strangulating intussusception. The high percentage of cases with diarrhoea may point in this direction. In addition, despite the long delay, the percentage of cases in which bowel resection was indicated, was not high compared to the literature (6-8,10-12).
Our low successrate of attempted hydrostatic reduction can be explained by three factors, the large proportion of older children, the long duration of symptoms and the high percentage of leading points. In children over two years of age lower hydrostatic reduction rates are encountered (13). Most intussusceptions become increasingly strangulated as time passes. Thus, when diagnosis and treatment are delayed, the likelihood of irreducibility increases. It is generally accepted that a high frequency of failures at barium enema reduction is related to the presence of a leading point (5,9). In children under two years of age we found a percentage of predisposing factors of 13 and over that age of even 39. These numbers also differ strikingly from those given in the literature (3,5). The smaller proportion of idiopathic intussusceptions is in accordance with the low incidence of intussusceptions under two years of age we calculated. We have no explanation for this low incidence.
From comparing our data with those previously reported in the literature we conclude that the group characteristics of children with intussusception can show important geographic variations.
We stress the importance of being aware of these possible variations in the light of early diagnosis and evaluating the results of treatment.

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III.2

Intussusception : Factors related to the outcome of treatment

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Abstract

In an effort to provide guide-lines on the choice of treatment in intussusception ten factors which are related to the outcome of therapy according to the literature, were studied in a series of 146 children with intussusception. A long duration of symptoms, vomiting, rectal bleeding, small bowel obstruction, ileoileocolic intussusception and the presence of a leading point were all significantly related to failure of attempted hydrostatic reduction. However only 'rectal bleeding' and 'a duration of symptoms of more than 48 hours' contributed significantly to predict failure of hydrostatic reduction according to logistic regression analysis. In our opinion besides the generally accepted contraindications, signs of peritonitis and of bowel perforation, the presence of rectal bleeding after a duration of symptoms of more than 48 hours is a contraindication for attempts at hydrostatic reduction. Archives of Disease in childhood (1990); intussusception, therapy.

Introduction

Hydrostatic reduction during barium enema has become an accepted method of management of intussusception in children. A success rate of over 80% can be achieved 1-3 . Controversy still exists concerning the contraindications for this non-operative treatment. Several authors administer the barium enema to every child, provided that the principles of a correct procedure are rigorously observed 4. Some, however, do not shrink away from increasing the pressure up to 150 cm water, bimanual manipulation and up to 10 attempts at hydrostatic reduction 1,3. The risk of bowel perforation during barium enema reduction is less than 1% 5,6. Resection because of irreducibility or non-viability is necessary in about 12% 7-11.

The presence of signs of peritonitis and of bowel perforation is considered to be an absolute contraindication for attempts at hydrostatic reduction. There is no agreement concerning the long duration of symptoms (over 24 or 48 hours) and evidence of small bowel obstruction as contraindications. Several other factors have been described to show a correlation with the outcome of treatment (Table I) 1,2,4-8,10-21. The diversity of opinion made us feel it worthwhile to review our series of intussusceptions. We submitted it to a statistical analysis with special interest for those intussusceptions that could not be reduced hydrostatically and those that required intestinal resection because of irreducibility or non-viability

at laparotomy. The aim of our study was to provide practical guidelines on the choice of treatment.

Methods

The fully documented records of 146 children under the age of 15 years, admitted to the Department of Pediatrics or (Pediatric) Surgery of the University Hospital St.Radboud and of the St.Canisius-Wilhelmina Hospital of Nijmegen from 1968 to 1988 in whom the clinical diagnosis of intussusception was supported by either radiological evidence or by laparotomy were reviewed. The data listed in Table I were recorded. Hydrostatic reduction was carried out by the method described by Ravitch 4. Absolute contraindications for an attempt at hydrostatic reduction were signs of peritonitis or bowel perforation. Other indications for primary surgical treatment were a bad general condition of the child, a duration of symptoms of more than 48 hours and pronounced small bowel obstruction. Since these criteria were not rigidly followed, it was possible to examine the outcome of the treatment against these criteria. Children with abdominal distension, hyper-resonance, abnormal bowel sounds, air-fluid levels and grossly distended bowel loops on the plain abdominal radiograph were considered to have pronounced small bowel obstruction. An intussusception was called ileoileo(coeco-)colic if evidence for an ileoileal component was found radiologically or at laparotomy.

As to the outcome of treatment four groups were distinguished. All children, successfully treated by hydrostatic reduction, formed group I. Included in this group were all cases in which after unsuccessful attempt at hydrostatic reduction the intussusception proved to be reduced at laparotomy (13 cases) or when the clinical course suggested reduction and no further therapy was necessary (1 case). Group II consisted of all cases treated by laparotomy and manual reduction after an unsuccessful attempt at hydrostatic reduction. The cases treated by primary laparotomy and manual reduction formed group III. All children treated by bowel resection because of irreducibility or gangrene formed group IV. The four groups were compared in three different ways. First the group successfully treated by hydrostatic reduction was compared to the group of children treated surgically (group I versus group II + III + IV). To exclude subjective influences that led to primary surgical therapy (group III), children, successfully treated by hydrostatic reduction, were also compared to children, treated surgically after unsuccessful attempt at hydrostatic reduction or treated by resection (group I versus group II + IV). Finally the group treated by resection was

compared to the children treated by hydrostatic reduction or by manual reduction only at laparotomy (group IV versus group I + II + III).
All univariate comparisons between different (sets of) groups where done with the usual X^2 test (size of test alpha = 0.05). A stepwise forward logistic regression analysis was used to assess which factor or combination of factors would have had the best prognostic power with regard to the outcome of treatment.

Results

Sixteen patients had an enteric intussuception. A predisposing factor precipitating intussusception was present in 12 cases. One jejunal and 6 ileal resections, 2 resections of a Meckel's diverticulum and 2 enterotomies were done. These enteric intussusceptions form a distinct group that should be managed surgically. They are not analysed here.
The remaining 130 children had colonic components of the intussusception and included 90 males and 40 females. Their ages ranged from one day to 14 years and 3 months. 25 Children were 6 months old or younger and 45 were over 3 years of age. The distribution of all patients according to the outcome of treatment is shown in Table 1. The distribution of possible treatment influencing factors over the four separate groups of patients is also shown in Table 1.

Table 1. Distribution of patients with factors related to the outcome of treatment of intussusception over 4 groups of different therapeutic modifications.

Factor related to outcome of treatment	Group I Hydrostatic reduction n=65 (%)	Group II Failed hydrostatic reduction + laparotomy n=36 (%)	Group III Primary laparotomy N=21 (%)	Group IV Bowel resection N=8 (%)
Age ≥36 months	19 (29)	9 (25)	4 (19)	2 (25)
Duration of symptoms ≥48 hours	13 (20)	19 (53)	6 (29)	4 (50)
Vomiting	45 (69)	29 (81)	21 (100)	8 (100)
Absence of abdominal pain	10 (15)	2 (6)	2 (10)	5 (63)
Rectal bleeding	11 (17)	17 (47)	14 (67)	6 (75)
Pronounced small bowel obstruction	7 (11)	9 (25)	10 (48)	7 (88)
White blood count ≥20 x 10.9/ml	2 (3)	3 (8)	1 (5)	2 (25)
Ileoileo(coeco)colic intusssusception	2 (3)	9 (25)	5 (24)	4 (50)
Apex at or beyond splenic flexure	7 (11)	6 (17)	4 (19)	4 (50)
Presence of leading point	4 (6)	7 (19)	6 (29)	5 (63)

Results of x^2 tests

Results of the X^2 tests are listed in table 2. These results were the same if different cutoff points were chosen for age and duration of symptoms.

Table 2. P-values of comparisons between different (sets of) groups according to X^2 test for factors related to the outcome of treatment of intussusception. n.s. = not significant (p>0.05). *1 value unknown. **12 values unknown.

Factor related to outcome of treatment	Group I versus Group II+III+IV	Group I versus Group II+IV	Group IV versus Group I+II+III
Age ≥36 months	n.s.	n.s.	n.s.
Duration of symptoms ≥48 hours*	0.003	0.001	n.s.
Vomiting	0.005	n.s.	n.s.
Absence of abdominal pain	n.s.	n.s.	0.000
Rectal bleeding	0.000	0.000	0.021
Pronounced small bowel obstruction	0.000	0.001	0.000
White blood count ≥20 x 10.9/ml**	n.s.	n.s.	0.034
Ileoileo(coeco)colic intussusception	0.000	0.000	0.019
Apex at or beyond splenic flexure	n.s.	n.s.	0.025
Presence of leading point	0.002	0.004	0.002

Stepwise logistic regression analysis

Group I versus group II + III + IV:

Only the factors "rectal bleeding" and "duration of symptoms >48 hours" contributed significantly to predict failure of attempt at hydrostatic reduction. Table 3 shows the results of the logistic regression procedure (classification using re-substitution method). Patients with rectal bleeding and a duration of symptoms of more than 48 hours had an estimated probability of failure of hydrostatic reduction of over 92%. According to the results 13.5% of all children successfully treated by hydrostatic reduction would have had a probability of failure of over 75%. 21% of all children treated surgi-

cally would have had a probability of hydrostatic reduction of over 78%.

Table 3. Result of logistic regression procedure for factors to predict failure of hydrostatic reduction.

	Predicted number of patients with	
	Failure of hydrostatic reduction	Success of hydrostatic reduction
Group I	12	46
Group II + III + IV	38	21

Sensitivity to detect failure of hydrostatic reduction 79.3%. Specificity 64.4%. Correct predictions 71.8%.

$\ln [p/(1-p)] = -1.3 + 2.4 X + 1.4 Y$
ln = natural logarithm
p = probability of failure of attempt at hydrostatic reduction
X = rectal bleeding, present = 1, absent = 0
Y = duration of symptoms > 48 hours, present = 1, absent = 0

Group I versus group II + IV:

The results of the stepwise logistic regression procedure were essentially the same as for Group I versus group II + III + IV.

Group IV versus group I + II + III:

With regard to bowel resection two factors showed significant predictive value according to logistic regression analysis. These factors are "pronounced small bowel obstruction" and "absence of abdominal pain" (Table 4). Patients with pronounced small bowel obstruction but without abdominal pain had a probability of bowel resection of over 82%. The 3% of all children successfully treated by hydrostatic reduction or manual reduction at laparotomy that were wrongly classified had a probability of bowel resection of 56%. In 3 of all 8 cases of bowel resection there was a probability of resection of 12% or less.

Table 4. Result of logistic regression procedure for factors to predict bowel resection.

	Predicted	
	Bowel resection	No bowel resection
Group IV	5	3
Group I + II + III	3	106

Sensitivity to detect bowel resection 62.5%. Specificity 97.2%. Correct predictions 94.9%.

$\ln [p/(1-p)] = -2.8 + 3.1 X + 2.2 Y$
ln = natural logarithm
p = probability of bowel resection
X = pronounced small bowel obstruction, present = 1, absent = 0
Y = absence of abdominal pain = 1, abdominal pain = 0.

Discussion

The use of barium enema reduction has gained acceptance as the initial procedure in most cases of intussusception. The reduction rates reported vary enormously 9. Low rates may be explained by a low incidence of intussusception and lack of experience. The use of premedication and a more vigorous reduction technique may lead to a higher reduction rate. However signs of peritonitis and of bowel perforation are generally accepted indications for primary surgical treatment.

Several authors have reported a lower hydrostatic reduction rate for children under 1 year of age 2,10,15. Higher resection rates were also reported for these children 14,15. Eklöf could not find an explanation in a higher incidence of leading points in older children. He assumed the ileocoecal valve in infants to be more competent, for unknown reasons. In children over three years of age lower hydrostatic reduction rates and a higher incidence of leading points are encountered 2,12,16,17. In contradiction to these studies and in confirmity with Gierup, Rosenkrantz and Liu we found the patients age not to be a significant factor as to the outcome of treatment.

Most intussusceptions become increasingly strangulated as time passes. Thus, when diagnosis and treatment are delayed, the likelihood of irreducibility increases. A short duration of symptoms favors successful hydrostatic reduction 1,6,9,12,15,16. Liu and Rosenkrantz proved this to be significant for a 48 hour cutoff point and Sparnon for a 12 hour cutoff point. However, Leonidas stated that the duration of symptoms does not seem critical in the decision

making proces as to what treatment should be chosen. In contradiction, in our material significantly fewer children with symptoms for more than 48 hours were in the group treated hydrostatically compared to the group that had surgical therapy. Several authors have reported resection rates of over 20% after 48 hours duration of symptoms 11,12,18. Although in 4 of 8 cases of bowel resection a duration of more than 48 hours was recorded, this was not significantly different from those cases in which no resection was necessary.

In the first instance vomiting in children with intussusception is reflexogen in origin. Later in the course of the disease vomiting is the result of bowel obstruction. Gierup found much lower hydrostatic reduction rates in cases with vomiting as the only or as a concurrent symptom. In our material vomiting was related to the outcome of attempts at hydrostatic reduction.

Sparnon and Ein reported much lower hydrostatic reduction rates in children with intussusception without abdominal pain. A good explanation for the relation between the absence of pain and the outcome of treatment has not been given. We could only confirm that significantly more painless intussusceptions were present in the group that needed resection.

Strangulation of intussuscepted bowel causes venous compression, swelling of tissue and discharge of mucus mixed with blood, currant-jelly stools. Gierup found a 57% hydrostatic reduction rate in cases with bloody stools opposed to 86% for cases without rectal bleeding. In Liu's material rectal bleeding was also associated with a low reduction rate, but it achieved no significance. However, in our material it did in every way.

In most intussusceptions there is a simultaneous interference with the vascular supply of the intussusceptum and with the patency of the alimentary canal. In accordance with our results several authors found a statistically significant relation between the presence of bowel obstruction and the complicated intussusception 6,8,13. Although many authors report a hydrostatic reduction rate of obstructed intussusceptions of well under 40% 5,6,12,16. others reported success rates of 50% or more 1,13. The definition of bowel obstruction may be of influence on these differences. There is a relation between the duration of symptoms and small bowel obstruction 8. The risk of perforation is increased, especially in young, sick infants who have a long history (> 36 hours) and small bowel obstruction 5.

Laboratory aids, such as white blood count, are usually of no great diagnostic benefit when predicting failure of attempts at hydrostatic reduction. On the other hand we did find a significantly high proportion of patients who needed resection and had a white blood

count of more than 20 x 10.9/ml with a shift to the left. This is in confirmity with a report from the Mayo Clinic 18.
Ileoileocolic intussusceptions form a special problem. Gierup found them associated with a high percentage of organic lesions. Potts had to do a resection in half the cases of ileoileocolic intussusception. As in our material the hydrostatic reduction rate was low. It is assumed that not much pressure can be exerted beyond the ileocoecal valve during barium enema. Probably the last part of the reduction of an ileoileocolic intussusception is for the greater part spontaneous. It requires a high degree of suspicion after hydrostatic reduction through the ileocoecal valve to detect an ileoileal component. As the type of intussusception is not known before a choice of treatment is made, it is of little clinical impact.
Nordshus found a much lower hydrostatic reduction rate for intussusceptions reaching beyond the transverse colon. Like several other authors we did not find a relation between the site of the apex and the hydrostatic reduction rate 1,3,12. In our material it was related to a significantly high percentage of resections. Many intussusceptions reaching far into the colon must have a wide ileocoecal valve or they must be of the ileo-coeco-colo-colic type in which the colic circumference acts as the relatively wide neck of the intussusception.
It is generally accepted that a high frequency of failures at barium enema reduction is related to the presence of a leading point. In the past indications have been found that intussusceptions caused by a leading point are unlikely to be reduced at all by hydrostatic barium enema and will almost certainly require surgical exploration 4,7. More recently exceptions to this statement have been described 21. Our results also show that hydrostatic reduction is possible in the presence of a leading point. However the reduction rate and the resection rate are significantly different. There is no clear anatomical or pathophysiological explanation for this phenomenon. This result is of little clinical importance as the presence of a leading point is not known in most cases.
The aim of our study was to provide guide-lines on the choice of treatment of children with intussusception, in which cases no attempt at hydrostatic reduction should be made and under which circumstances it should be attempted very cautiously. To reach such recommendations one has to decide which probability of mortality and morbidity as well as which delay and associated need for bowel resection are acceptable. According to Leonidas survival seems to be less critical since mortality is very low. He calculated for morbidity that hydrostatic reduction is the best therapeutic option if the anticipated rate of success exceeds 14% 6. The delay

caused by an attempt at hydrostatic reduction has never been evaluated but it does not seem very important.
In conclusion in our opinion no attempt at hydrostatic reduction should be made in the presence of rectal bleeding after a duration of symptoms of more than 48 hours. If in the presence of rectal bleeding an attempt at hydrostatic reduction is made within two days, there is a probability of more than 25% of success. We calculated a probability of almost 50% of hydrostatic reduction if the attempt was made after more than 48 hours in the absence of rectal bleeding. In these situations an attempt at hydrostatic reduction seems justified, provided the principles of a correct procedure are observed. The height of the reservoir should not exceed 100 cm and the abdomen should not be manipulated. In the absence of rectal bleeding and after a duration of symptoms of less than 48 hours it seems justified to always make an attempt at hydrostatic reduction.

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III.3

Intussusception : "Neurological" signs as early manifestations.

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Abstract

The appearance of neurological signs early in the course of intussusception is a not well-understood phenomenon, not adequately stressed in the literature. The records of 148 children with intussusception were examined in order to study a possible correlation of these neurological signs with other characteristics of intussusception. We found a significant correlation with a duration of symptoms less than 48 hours, rectal bleeding, bowel obstruction, intussusception with ileoileal component, dehydration, bicarbonate <20 mmol/l and hydrostatic irreducibility. The origin of these signs, toxic metabolic, behavioral, reflexogen or sensory in nature, is discussed.

Introduction

Usually intussusception is a readily diagnosed disease as the clinical pattern of colicky abdominal pain, vomiting, bloody stools and an abdominal mass is well recognized. In recent reports the appearance of signs such as apathy, lethargy, altered consciousness and hypotonia early in the course of the disease has been emphasized.1-11 In the absence of most of the cardinal symptoms of intussusception the domination of the clinical picture by these early signs constitutes a diagnostic challenge to the clinician. These signs are often taken for neurological abnormalities and this results in intensive studies to eliminate known causes of central nervous system disturbances.
An adequate explanation for these early signs has never been given. The absorption of toxic metabolic products from a strangulated bowel 1-3, the release of neuropeptides 10,11, a reflexogen nature 12 or a behavioral reaction may be suggested.
In order to study a possible correlation with other characteristics, revealing the origin of these neurological signs, we examined the records of 148 children with intussusception.

Patients and methods

The records of 148 children under the age of 15 years, admitted to the Department of Pediatrics or Pediatric Surgery of the University Hospital St.Radboud and of the St.Canisius-Wilhelmina Hospital of Nijmegen from 1968 to 1988, in whom the clinical diagnosis of intussusception was supported by either radiological evidence or by laparotomy, were reviewed.

We recorded age, duration of symptoms, the presence of vomiting, abdominal pain or rectal bleeding, body temperature at admission, signs of dehydration or bowel obstruction, hematocrit, blood urea nitrogen, bicarbonate, white blood count, type of intussusception, location of the apex and outcome of treatment.
The patients were divided into 3 groups depending on the absence or presence of neurological signs at presentation (concomitant or dominant). Lethargy and somnolence associated with one or more of the cardinal symptoms of intussusception was considered concomitant. Lethargy and somnolence, associated with unresponsiveness and generalized hypotonia, overshadowing abdominal symptoms was considered dominant. Lethargy and somnolence in the absence of abdominal symptoms was also called dominant.
As to the outcome of treatment 4 groups were distinguished. All children, successfully treated by hydrostatic reduction, formed group 1. Included in this group were all cases in which after unsuccessful attempt at hydrostatic reduction the intussusception proved to be reduced at laparotomy (13 cases) or when the clinical course suggested reduction and no further therapy was necessary (1 case). Group 2 consisted of all cases treated by laparotomy and manual redcution after an unsuccessful attempt at hydrostatic reduction. The cases treated by primary laparotomy and manual reduction formed group 3. All children treated by bowel resection because of irreducibility or gangrene formed group 4. Children with abdominal distension, hyperresonance, abnormal bowel sounds, air-fluid levels and grossly distended bowel loops on the plain abdominal radiograph were considered to have pronounced small bowel obstruction. An intussusception was called ileoileo(coeco)colic if evidence for an ileoileal component was found radiologically or at laparotomy. Dehydration was a clinical diagnosis based on the presence of diminished turgor, dry mucous membranes and sunken fontanelles.
All univariate comparisons between different (sets of) groups were done with the usual X^2 test (size of test alpha = 0.05).
The cut off points for age were chosen at 6, 12 and 24 months. The duration of symptoms was analysed at 24 and 48 hours. The cut off point for body temperature at admission was 38°C.
Laboratory findings were studied for the whole group at fixed cut off points (white blood count at 10 x 10.9/l and at 20 x 10.9/l, hematocrit at 0.40, blood urea nitrogen at 7 mmol/l and bicarbonate at 20 mmol/l). Besides, pairs of patients, matched for age, duration of symptoms, type of intussusception and localisation of the apex, were compared for laboratory findings at cut off points in accordance to their age. 13

Results

Statistical analysis failed to show a significant correlation between on the one site the presence of neurological signs and on the other side patients age, the presence of vomiting and of abdominal pain, body temperature at admission, raised hematocrit or blood urea nitrogen, leucocytosis, the location of the apex and the necessity of bowel resection.
Table 1 shows the distribution of patients with and without neurological signs over those factors which were significantly correlated.

Table 1. Significant correlations of neurological signs and other characteristics in 148 cases of intussusception.

Characteristic of intussusception	Neurological signs Absent n = 92	Concomitant n = 44	Dominant n = 12	P-value
Duration of symptoms < 48 hours	58	24	12	0.01<P<0.05
,, ,, ,, ≥ 48 hours	34	20	0	
Rectal bleeding present	24	23	4	0.01<P<0.05
,, ,, absent	68	21	8	
Bowel obstruction present	21	19	2	0.01<P<0.05
,, ,, absent	71	25	10	
Intussusception with ileoileal component	17	17	2	0.01<P<0.05
Intussusception without ileo-ileal component	75	27	10	
Dehydration present	16	21	6	P<0.001
,, absent	76	23	6	
Bicarbonate < 20 mmol/l	35	20	9	0.01<P<0.05
,, ≥ 20 mmol/l	57	24	3	
Treatment group 1	48	12	6	0.01<P<0.05
,, group 2+3+4	43	31	6	
Treatment group 1	48	12	6	0.01<P<0.05
,, group 2+4	28	20	4	

Comment

The clinical pattern of intussusception is typical but a complete picture is only present in approximately 10% of cases. It is therefore

not surprising that over 40% of patients are first hospitalized up to 24 hours after the onset of symptoms. Atypical presentations with marked variations from the classic picture as well as diagnostical errors have been described as obstacles to early diagnosis. 5 However, early diagnosis is important because there is a close relationship between the duration of intussusception and the outcome of attempts at hydrostatic reduction and at manual reduction during laparotomy. 14

The association of certain alterations in neurological status such as restlessness, lethargy, altered consciousness, stupor or convulsions, has been mentioned occasionally in conjunction with intussusception. In the majority of cases these signs were late manifestations due to fluid and electrolyte imbalance, sepsis or blood loss following bowel obstruction or necrosis. 8,15,16 However it has long been known that neurologic signs and symptoms are commonly seen early in the course of this disorder. Oberhelman and Condon reported that shock or impending shock as an early symptom is not adequately stressed in the literature. 17 Thurston concluded that a "knocked out" appearance was highly characteristic of the early stages of acute intussusception. 18 We found a significant correlation between neurological signs and a duration of symptoms of less than 48 hours. It is most likely that the presence of neurological signs prompted the family doctors to early admission.

In more recent years several cases have been reported in which neurological manifestations such as altered consciousness, twitches and hypotonia dominated the more common features or were the only early features. 2-10 One well documented series has been reported. 1 As in our material in the above mentioned cases no common predisposing cause, anatomical variation or enviromental circumstance to explain the neurological status, was found. A cause and effect relationship between the intussusception and altered consciousness and hypotonia is strongly suggested. These neurological signs have been explained by absorption of some metabolic product from an ischemic bowel. 1-3 The significant correlation we found between the neurological signs and the presence of bloody stools and bowel obstruction suggests the influence of strangulation of the bowel on the neurological status. This is supported by the significant correlation between the neurological signs and intussusception with an ileoileal component. These intussusceptions are often strangulated in children. On the other hand this explanation for a phenomenon, often so early in the course of the disease, seems less likely, the more so as we did not find any non-viable bowel at laparotomy in the cases with the most serious neurological signs. This is in accordance with most cases reported in the literature. In addition, a normally functioning liver eliminates all toxic

metabolic products from the portal venous blood, preventing these products to pass the blood brain barrier. On the contrary, successful attempts at hydrostatic reduction, an indication of little strangulation, were significantly related to the presence of neurological signs.

In our material dehydration, found at physical examination, showed to be significantly more frequent in patients with neurological signs. However this was neither supported by an important correlation with a high hematocrit nor with azotemia. Azotemia was found by Heldrich in only 2 of 19 lethargic patients. 1 Azotemia, of course, is less likely to occur in children with low protein intake due to vomiting and anorexia. In addition we found a high percentage of cases with severe neurological signs with evidence of acidosis. This did not account for the group with less severe signs.

The altered consciousness and hypotonia might be a normal behavioral reaction of a child at the age of about 1 year to an attack of severe abdominal pain. On the other hand they might be reflexogen in nature. 12 In children under the age of 1 year syncope or seizures provoked by several different stimuli have been described. 12,18 Severe pain must be capable of doing so but the exact mechanism is unknown. De Lorimier stated that the release of endorphins and enkephalins following stress, especially pain, might play a role. He suggested that profound lethargy, somnolence or even semicoma were a manifestation of pain. 11 Tenenbein speculated that an endogenous opioid like met-enkephalin could induce coma and mask the abdominal pain in intussusception. 10 Endorphin and (met-)enkephalin belong to a group of peptides which are found both in the brain and in the gastrointestinal tract. Several of these brain-gut peptides have been demonstrated in primary sensory neurons, in sensory neurons of the dorsal horn of the spinal cord and in areas in the brain associated with the sensory input of pain signals 20, 21. It is unclear wether one of the factors correlated with the presence of neurological signs is also related to more or more severe pain.

Whatever the cause, intussusception should be considered in previously healthy children with apathy, lethargy, hypotonia and sudden alteration of consciousness even in the absence of classic signs when known conditions of altered consciousness have been systematically excluded. This will hasten diagnosis and therapy and lessen morbidity.

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III.4

Chronic intussusception in children

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Abstract

Nine children presented with intussusception lasting for 14 days or more. Their mean age was 8.5 years. Diagnosis of intussusception was delayed considerably, probably due to an unusual presentation. Compared with acute intussusception, symptoms consist of infrequent attacks of abdominal pain, sporadic vomiting and no, or small changes in defecation. Marked weight loss and an abdominal mass assume diagnostic significance, in contradiction to bloody stools. Ultrasonography can be of diagnostic value. An attempt at hydrostatic reduction is often unsuccessful. A high frequency of organic lesions precipitating intussusception warrants early surgical intervention.
Keywords: Intussusception, children.

Introduction

Intussusception is considered to be a readily diagnosed disease because the classical clinical pattern of colicky abdominal pain, vomiting and bloody stools in a child under two years of age is well recognized. However the fully developed triad of cardinal symptoms has been noticed in less than 50 per cent of cases (1, 2). The presence of symptoms depends on the varying degree of strangulation of the bowel.
In the absence of strangulation currant-jelly stools and vomiting (due to intestinal obstruction), abdominal distension, abnormal bowel sounds and constipation may be absent. A non-strangulating intussusception may reduce spontaneously if not protruded too far or strangulate if it progresses. It may also stay unchanged for several days or even months. In the past intussusceptions lasting for 14 days or more have been considered to be chronic (4, 5, 6). They are distinguished from acute forms of intussusception by a characteristic clinical pattern that is conducive to delay in diagnosis. This paper reports the clinical pattern in and diagnostic features of nine cases of chronic intussusception in children.

Patients and methods

The records of all 139 patients under the age of 15, admitted to the Department of General Surgery of the St. Canisius-Wilhelmina Hospital or to the Paediatric Surgical Centre of the St. Radboud Hospital of the Catholic University of Nijmegen from 1968 to 1988 in whom the clinical diagnosis of intussusception was supported

either by radiological evidence or by laparotomy, were subject to a retrospective study. Nine patients had a chronic intussusception as defined above.

Results

The mean age was almost 8,5 years for the 7 male and 2 female patients (range from 3 years and 3 months to 14 years and 2 months). Three had an intussusception of the enteric type and three were ileo-ileo-colic intussusceptions. Two were of the ileo-caecal type and there was one appendico-caeco-colic intussusception. In 6 cases a leading point was found. Every day all 9 patients had complaints of intermittent abdominal pain. In 8 of 9 cases the frequency of the attacks was 5 times a day or less. 7 Patients vomited (also sporadically). Two patients lost blood per anum. One patient complained of constipation. In three cases a looser stool or diarrhoea was produced at a normal frequency. In five cases there was no change in defecation. 6 Patients were anorectic. At physical examination the weight of 3 patients was below the 10th percentile and in 5 cases between the 10th and 50th percentile. Signs of intestinal obstruction, abdominal distension and abnormal bowel sounds were found in 4 cases. An abdominal mass was present in 7 patients.
The mean time between the onset of symptoms and admission was 35 days ranging from 3 to 93 days. A chronic intussusception was suspected because of the continuing presence of symptoms and ill health of all children. In 3 cases there was a considerable delay in diagnosis of 7, 9 and 13 days after admission. In the last two of these cases diagnosis at admission was appendicular mass. In 8 cases diagnosis was reached pre-operatively: by clinical presentation (2 cases), by barium enema examination (3 cases), by small bowel follow-through meal (1 case) and by ultrasonography (2 cases). A plain abdominal radiograph of 3 patients showed air-fluid levels in dilated bowel loops on the day the diagnosis of intussusception was reached.
All children were operated on. An attempt at hydrostatic reduction was made in 4 cases but without succes. In three of these cases an organic lesion was found. At laparotomy the fourth case was found to have undergone reduction. Although, retrospectively, an intussusception could also be seen on an earlier barium enema, this was the only case were a recurrent acute intussusception could not be ruled out with certainty. In four patients manual reduction was performed easily, in one case followed by enterotomy and in another by a small resection of vital jejunum because of a tumor which turned out to be a fibroid polyp. In two cases with a duration of 93

and 56 days manual reduction could only be achieved with great difficulty because of adhesions after which ileocaecal resection was done because of questionable viability. Partial ileal resection without manual reduction, because of possibly non-viable bowel, was performed in two cases with a duration of 16 and 21 days.

Discussion

Strangulation of the bowel can occur early or late in the course of an intussusception. An intussusception lasting more than 14 days is an uncommon condition. The incidence is estimated at 5 per cent of all intussusceptions. Under the age of two years, when most acute intussusceptions happen, an incidence of 3 per cent has been calculated while over that age it was 10 per cent (7). This suggests that at an older age the anatomical conditions allowing an intussusception to occur without any significant impairment of the blood supply are more likely to be present. On the other hand the possibility remains that delay in diagnosis, caused by the older age of the child, permits a non-strangulating intussusception to last more than 14 days.

Chronic intussusceptions distinguish themself from acute intussusceptions by less severe abdominal pain and vomiting, a lower percentage of rectal bleeding and a higher percentage of diarrhoea (1-3, 5, 7). A striking feature is the significant weight loss in many children with chronic intussusception due to the longstanding anorexia and nausea or to the underlying causative factor involved (5, 7).

Besides small bowel follow through and barium enema, especially if a mass is found, ultrasonography can be of diagnostic value (5, 8-11). The sonographic picture of intussusception is rather characteristic. A cross section through the apex shows a target-like pattern. Scanning through the more proximal intussuscepted bowel demonstrates an image composed of two concentric rings and an inner circular area. Scans along the longitudinal axis exactly through the centre of the intussusceptum show four parallel stripes of low echogenity, delineating three reflective areas. Longitudinal scans not through the centre of the intussusceptum demonstrate three stripes of low echogenity seperated by two hyperreflective stripes.

Attempts at hydrostatic reduction of chronic intussusceptions have not been very successful in the past (5, 12). The high incidence of organic lesions is another reason for early surgical intervention, the more so as in older children this organic lesion frequently turns out to be a lymphosarcoma (12). At laparotomy manual reduction can

often be done with ease (5, 12). In case of irreducibility, questionable viability of the bowel and suspicion of intramural pathology, resection is indicated.

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III.5

Intussusception in children 5-15 years of age

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Summary

Twenty cases of intussusception in children between the ages of 5 and 15 years were compared with intussusception in infancy and early childhood. They accounted for 18.5 per cent of all 108 children treated for intussusception in two large hospitals from 1964 to 1984. Diagnosis of intussusception was delayed, probably due to an unusual presentation. Fifty-five per cent had a definite predisposing factor precipitating the intussusception and 45 per cent had a small bowel intussusception, which warranted early surgical intervention. In the absence of contraindications no child should be disqualified from an attempt at hydrostatic reduction. After hydrostatic reduction careful follow up is required to exclude an organic lesion, possibly by a small bowel follow-through meal. Surgery is indicated after hydrostatic reduction in case of chronically recurrent abdominal complaints.
Keywords: Intussusception, children.

Introduction

Intussusception is the commonest cause of small bowel obstruction in children between the ages of 2 months and 5 years. After the first year the incidence gradually declines. Intussusception is uncommon in children at the age of 5 years or over (1-4).
Whereas intussusception in children under the age of 5 years has often been the subject of studies, little attention has been paid to intussusception in children 5-15 years of age. This prompted the authors to examine the records of all 108 children treated for intussusception over a 20-year period in our two hospitals serving a district of 300 000 people. Children between the ages of 5 and 15 years represented 18.5 per cent of the total.
In the records of the 20 patients in this age group we found several striking differences when compared with recently published reports of large series which considered predominantly children under the age of 5 years (4-9).

Patients and methods

The records of all 108 patients under the age of 15, admitted to the Department of General Surgery of the St. Canisius-Wilhelmina Hospital or to the Paediatric Surgical Centre of the St. Radboud Hospital of the Catholic University of Nijmegen from 1964 to 1984 in whom the clinical diagnosis of intussusception was supported

either by radiological evidence or by laparotomy, were subject to a retrospective study. Twenty patients were between the ages of 5 and 15 years. The records of the paediatrician and/or (paediatric) surgeon as well as the available X-rays were reviewed.

Results

The mean age was 8,5 years for the 10 male and 10 female patients. Nine had an intussusception of the enteric type (jejuno-jejunal or ileo-ileal), one was an ileo-ileocolic intussusception, and the rest were of the ileo-(ceco-)colic type. In only two cases was the classic triad of colicky pain, vomiting and rectal bleeding present (Table 1). Six children showed only one symptom: pain or vomiting. A palpable abdominal mass was present in four of five patients whose sole complaint was abdominal pain; nine of the eleven children with colicky pain and vomiting but without bloody stools had a palpable mass. In only five patients was the diagnosis made within 24h after the onset of symptoms, while eight had had symptoms for more than 3 days. With the exclusion of three chronic intussusceptions the correct diagnosis was reached on an average of 3 days after the onset of symptoms. Five children had had recurrent abdominal complaints for a period of 8, 8, 6, 6, and 3 months respectively. In 14 the diagnosis was made clinically and by barium enema examination. Six cases were diagnosed at laparotomy.

Table 1. Clinical features.

Symptoms	Numbers of cases (n= 20)	Cases with abdominal mass
Pain	5	4
Pain + vomiting	11	9
Vomiting	1	-
Pain + rectal bleeding	1	-
Pain, vomiting + rectal bleeding	2	1

Treatment.
An attempt at hydrostatic reduction was performed in 11 children and was successful in 6. In three of the five cases of failed hydrostatic reduction a predisposing factor precipitating intussusception was found. Gangrene necessitated a partial resection of the small

bowel in the fourth patient. During laparotomy in the fifth child the intussusception was found to have undergone spontaneous reduction.

Laparotomy was performed on 14 patients. One intussusception reduced spontaneously, and manual reduction was performed in 13 cases. Enterotomy was carried out for excision of a polyp (2), for biopsy of marked lymphoid hyperplasia (1) and for biopsy of an ulcer in the caecum (1). Partial jejunal resection (10) was performed because of a tumour which turned out to be a fibroid polyp. On four occasions partial resection of the ileum was required because of non-viable bowel.

Out of the group of 14 children undergoing laparotomy a definite predisposing factor precipitating the intussusception was present in 11 cases (Table 2). In all three children with chronic intussusception and in four of the five with recurrent abdominal complaints laparotomy revealed an organic lesion.

Table 2. Predisposing factors in 20 cases of intussusception in children 5-15 years of age.

Aetiological factor	Number of cases
Meckel's diverticulum	2
Peutz-Jeghers syndrome	2
Appendiceal stump	1
Hamartoma	1
Fibroid polyp	1
Lymphoid hyperplasia	1
Mucoviscidosis	1
Severe nephrotic syndrome	1
Postoperative phase correction hernia hiatus oesophagi	1
Total	11

Discussion

In recently published reports from large children's hospitals the 5-15 age group account for less than 5 per cent (2-4), compared with

18.5 per cent in this series. This relatively high percentage is probably not due to a higher incidence but to a lower incidence of intussusception in infancy and early childhood. We calculated a low incidence of intussusception of 1.1 per 1000 live births compared with incidences of 1.57 to over 4 per 1000 live births reported in a review by Ravitch (1). Per year we saw one patient between the ages of 5 and 15 years with intussusception. Intussusception is thus an uncommon cause of abdominal complaints in the older age group (10).

The striking differences in presentation is conducive to delay in diagnosis (10). In children under the age of 5 years the classic triad of pain, vomiting and rectal bleeding is present in about 20-50 per cent (7,11). At school age we found it to be less frequent (10 per cent). Abdominal pain (in 75-94 per cent) and vomiting (in 63-92 per cent) are by far the most common presenting features in intussusception (1,5-9). In infants and younger children rectal bleeding is a rather frequent symptom (46-67 per cent) (4-8). We found rectal bleeding to be rare. In contrast an abdominal mass, usually found in 50-85 per cent (1,4-8), is a significant diagnostic finding. An abdominal mass was present in 14 of our patients. To avoid delay in diagnosis we recommend barium enema examination in any child who might possibly have an intussusception. In experienced hands ultrasonography can be most helpful in evaluating atypical cases (12).

In infancy and childhood intussusception is due to an organic lesion in 2-12 per cent (1,4-9), while in the older child there is a much higher incidence (3,6-8). Intussusception due to organic lesions is more difficult to reduce hydrostatically (9). The enteric or ileo-ileocolic type is more common in the older child (8,9), and this type is difficult to treat by hydrostatic reduction (9). It is the general experience that there is a higher failure of hydrostatic reduction in older children (10). Eklof (1980) and Jennings (1984) reported a more than 20 per cent overall success rate (2,3). In our view older children can be treated by hydrostatic reduction provided there is adequate follow-up and an organic lesion of the small bowel has been excluded. In the case of chronically recurrent abdominal complaints surgery is indicated.

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III.6

Diagnosis and treatment of adult intussusception

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Summary

Twenty adults were treated for intussusception in two large hospitals from 1969 to 1988. Fourteen intussusceptions originated in the small intestine and six in the large intestine. Diagnosis was reached pre-operatively in only 10 patients, probably due to the atypical clinical picture. In addition to a high degree of suspicion, careful examination of plain abdominal radiograph and ultrasonography are helpful in diagnosing adult intussusception. In 18 out of 20 patients, an organic lesion causing intussusception was found. In six patients the cause was a malignancy. In such cases surgical treatment is necessary. In jejuno-jejunal and ileoileal intussusceptions, an attempt at primary reduction followed by resection or enterotomy is justified. In most cases of ileocolic, ileo-cecocolic, and colocolic intussusception primary resection is the treatment of choice, especially in patients over 60 years old because of a high incidence of malignancy.
Keywords: Intussusception, adult, diagnosis, treatment

Introduction

In adults diagnosis of intussusception is usually delayed and not made before laparotomy. In contrast to childhood intussusception the clinical picture in adults is not typical and not well-recognized. Moreover, the incidence of adult intussusception is low [1-8]. It constitutes approximately 5% of all intussusceptions and intussusceptions account for 5% of all cases of intestinal obstruction [3,4,7]. Most surgeons have a limited experience with the clinical entity and with the various considerations involved in the diagnosis and management.

We examined the record of 20 adults treated for intussusception over a 20-year period in our 2 hospitals serving a district of 300,000 persons. Having compared our data with reviews and recently published well-documented reports of large series concerning adult intussusception, it appears that diagnosis and treatment of adult intussusception ask for a specific approach (1-10).

Material and methods

The records of 20 patients over 15 years of age admitted to the department of general surgery of the St. Canisius-Wilhelmina Hospital in Nijmegen and of the St. Radboud Hospital of the Catholic University of Nijmegen from 1969 to 1988, in whom the diag-

nosis of intussusception was documented at laparotomy, were retrospectively studied. Patients with postoperative intussusceptions, intussusceptions caused by intestinal intubation, agonal intussusceptions, rectal or stomal prolapses, jejuno-gastric intussusceptions after gastro-enterostomies and intussusceptions incidentally noticed at laparotomy were not included in this study.
There were 12 men and 8 women in this series. Their ages ranged from 15 to 81 years with a mean age of 39 years. Only one patient was under 18 years of age. Five cases were of the jejuno-jejunal type, 6 of the ileo-ileal type and 3 of the ileo-ileo-colic type bringing the total of intussusceptions originating in the small intestine up to 14. Five patients had an ileo-ceco-colic intussusception and one a sigmoido-rectal intussusception.

Results

The presenting clinical picture of at least 15 patients was not typical (Table 1). Fifteen patients presented within 10 days after the onset of symptoms but five had chronically recurrent abdominal complaints. In five cases symptoms were chronic, extending backwards from 1 month to 5 years. Fourteen patients showed signs of partial or complete intestinal obstruction. Colicky pain and vomiting as well as an abdominal mass were present in 7 of the 10 patients in whom diagnosis was reached pre-operatively by clinical presentation and plain abdominal radiography (4 patients), barium enema examination and sigmoidoscopy (1 patient), small bowel follow-through meal (2 patients) and ultrasonography (3 patients). During the last 4 years of the examination period ultrasonography was liberally applied in case of suspicion of intussusception. In retrospect a plain abdominal radiograph of eight patients showed signs strongly suggestive of intussusception: a soft tissue mass in six patients, air-fluid levels in dilated bowel loops in six patients, and scant colonic gas and fecal content in six patients.
In 18 patients, a causative organic lesion was found (Table 1). In the other two patients, pregnancy and adhesions after partial gastric resection may have been predisposing factors. In three cases of Peutz-Jeghers syndrome reduction and multiple enterotomies for polypectomy were performed. In 10 patients with small intestinal intussusceptions, partial resection was performed because of a palpable pathological process after manual reduction in 4 patients and irreducibility in 6 patients. In two of the cases of small intestinal intussusception, a lymphosarcoma was found. For all 5 ileo-ceco-colic intussusceptions resection was performed because of irreducibility and a palpable pathologic process which in four cases

Table 1. Clinical features in 20 cases of adult intussusception*

	Intussusception	
	Small intestinal (n= 14)	Large intestinal (n= 6)
Symptoms		
Pain	2 (0)	1 (1)
Pain & Vomiting	8 (5)	2 (0)
Pain & Rectal bleeding		1 (1)
Pain, vomiting & rectal bleeding	3 (3)	1 (1)
Asymptomatic	1 (0)	1 (1)
Causative lesion		
Meckel's diverticulum	3	
Peutz-Jeghers syndrome	4	0
Fibrolipoma	1	0
Fibroid polyp	1	0
Lymphoid hyperplasia	1	0
Lymphosarcoma	2	1
Adenomatous polyp	0	1
Adenocarcinoma	0	3

* Numbers in parentheses indicate patients with an abdominal or rectal mass.

turned out to be a malignancy. This was the case in all four patients over 60 years old. The one sigmoido-rectal intussusception was treated by reduction and low anterior resection after sigmoidoscopy failed to demonstrate a malignant process. The lead point turned out to be an adenomatous polyp. The case was complicated by an abscess in the pouch of Douglas. No other direct postoperative complications occurred. One patient had to be re-operated for adhesions 1 year after partial ileal resection. At last follow-up, there was no recurrence. Two patients died from the primary disease, a lymphosarcoma, 47 and 79 days after operation. All other patients with a malignancy were alive without evidence of disease 2, 2, 9 and 13 years after operation.

Comments

In Western countries, intussusception is rarely considered in the differential diagnosis of gastrointestinal disease in the adult. The presenting clinical picture is that of partial intestinal obstruction and resembles several other more common diseases. In view of the nonspecific symptoms of pain and vomiting and the infrequency of bloody stools the finding of an abdominal or rectal mass assumes diagnostic significance [1-3,7,9]. In many cases a plain abdominal radiograph will be made, and careful observation will facilitate early diagnosis [5-7,11,12]. As in children, abnormal findings in ileocolic and ileo-cecocolic intussusceptions are a soft tissue mass, a gas-outlined apex, air-fluid levels in dilated bowel loops, and scant colonic gas and fecal content (Figure 1).

Fig. 1. Plain abdominal radiograph of an adult patient with intussusception showing a soft tissue mass (arrows), a gas-outlined apex (arrowheads), air-fluid levels in dilated bowel loops and scant colonic gas and fecal content.

In case of suspicion of intussusception, especially if a mass is found, ultrasonography can be of great diagnostic value. The sonographic features of intussusception are rather characteristic. A cross-section through the apex of the intussusceptum shows a targetlike pattern. More proximal scanning demonstrates an image composed of two concentric rings and an inner circular area (Figure 2).

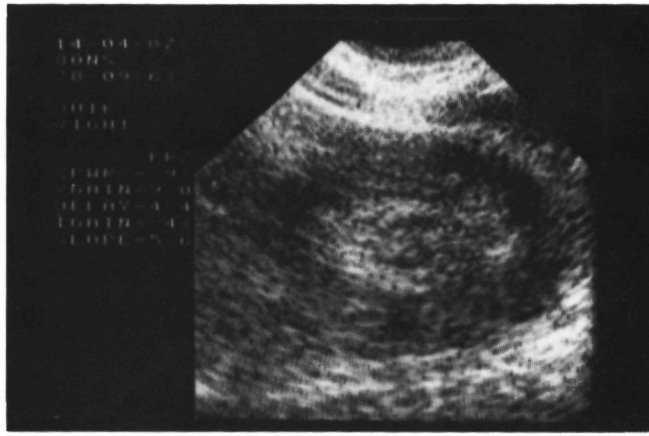

Fig. 2. Sonogram of the same patient as in Figure 1 showing a typical multiple concentric ring sign.

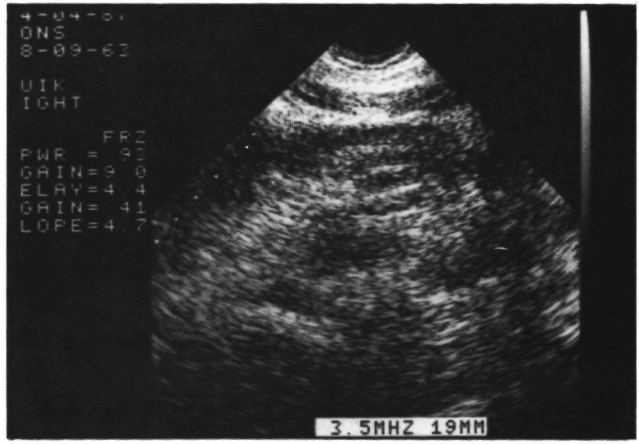

Fig. 3. Sonogram of the same patient as in figures 1 and 2 showing four parallel stripes of low echogenity seperated by three hyperreflective stripes.

Scans along the longitudinal axis exactly through the centre of the intussusceptum show four parallel stripes of low echogenity, delineating three reflective areas. Longitudinal scans not through the centre of the intussusceptum demonstrate three stripes of low echogenity separated by two hyperreflective stripes (Figure 3) [13,14].

In case of any doubt barium enema, endoscopy, and small bowel follow-through meal are suitable to confirm the diagnosis [5-7,9,11]. Other diagnostic tools such as angiography, technetium-99m pertechnetate scanning, and computed tomography can usually be omitted.

Intussusception in the adult is associated with a causative lesion in 75 to 90 percent of cases [2,4,5,7,9]. As in our series, neoplasms account for about 70 percent of adult intussusceptions, and in about one-third of all cases, the cause is a malignancy [1-7,9,10]. Some investigators have reported an even larger proportion [3,8,9]. As in the present study, the incidence of malignancy in intussusception in patients over 60 years of age is high, 50 to 70 percent has been reported [4,8,10]. Colocolic and ileo-cecocolic intussusceptions are associated with malignancy in 54 to 77 percent of cases [3-9]. Jejuno-jejunal and ileoileal intussusceptions are associated with malignancy in about one-fourth of cases [7-10]. According to some reports this malignancy is a metastasis in the majority of cases [9,15].

Treatment of adult intussusception must be surgical [1,4,9]. Intraoperative reduction might permit a more limited resection or avoid resection but there is a risk of perforation of ischemic bowel and anastomosis in a more edematous area. The danger of reduction of externally vital bowel with mucosal necrosis, intraluminal seeding, or venous embolization of malignant cells has been pointed out [5,9]. Jejuno-jejunal and ileoileal intussusceptions can be treated by gentle primary reduction and small resection or enterotomy as is the case in patients with Peutz-Jeghers syndrome [7,9]. In patients with a history of previous malignancy and possibly a metastasis leading the intussusception, primary reduction is considered not to influence long-term prognosis [9]. In patients over 60 years of age, no attempt at manual reduction should be made and primary resection is indicated. Colocolic, ileocolic and ileo-cecocolic intussusceptions should also be treated by primary resection with few exceptions [1,7,9]. In patients with sigmoido-rectal intussusception without endoscopically assessed evidence of malignancy, initial reduction may allow anterior resection or a two-stage resection [1,9].

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III.7

Intussusception complicated by Bowel Perforation during hydrostatic Reduction.

Invagination kompliziert durch Perforation des Colons während hydrostatischer Reposition.

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Summary

Most perforations of the bowel during attempt at hydrostatic reduction of intussusception occur in an area of localized infarction in the normal transverse or left colon. An animal model of intussusception was used to find indications for the cause of this phenomenon. We submitted the intussuscipiens of 10 strangulated intussusceptions in 6 dogs to a histological examination. In 6 of 10 intussusceptions we found ischemic changes in the mucosa of the intussuscipiens. In 3 cases these lesions were multiple. All lesions were found on locations where there was a close contact between the intussusceptum and the intussuscipiens. We did not find signs of impaired circulation of the whole intussuscipiens. We conlude that our findings give an indication that perforation of the intussuscipiens during attempt at hydrostatic reduction occurs through areas of localized ischemic infarction on the basis of direct pressure by the intussusceptum.
Key words: Intussusception – complication – pathology.

Invagination kompliziert durch Perforation des Colons während hydrostatischer Reposition.

Zusammenfassung

Den meisten Darmperforationen während hydrostatischer Reposition einer Invagination liegt ein lokal infarziertes Gebiet im Bereich des vitalen Colon transversum oder descendens zugrunde.
Wir suchten anhand eines Tiermodels nach Hinweisen für die Ursache dieses Phänomens. Das Intussuscipiens von 10 strangulierten Invaginationen wurde einer histologischen Untersuchung unterworfen. In 6 Fällen wurden ischämische Veränderungen der Schleimhaut des Intussuscipiens gefunden. In 3 Fälle handelte es sich um multiple Läsionen. In alle Fällen bestand ein enger Kontakt zwischen Invaginat und Intussuscipiens an der Stelle der ischämischen Veränderungen. Zeichen einer reduzierten Durchblutung der Intussuscipiens-Wand wurden nicht gefunden. Unsere Schlussfolgerung ist, dass unsere Befunde Hinweise darauf geben, dass Perforationen des Intussuscipiens während hydrostatischer Repositionsversuche an Stellen mit lokaler ischämischer Infarzierung stattfinden die durch direkten Druck des Invaginats verursacht werden.
Schlusselwörter: Invagination – Komplikation – Pathologie.

Introduction

Fortunately perforation of the bowel during attempt at hydrostatic reduction of intussusception is uncommon. The incidence is under 1% (7,9). Recently in our clinic we encountered the first perforation during hydrostatic reduction in a series of 105 attempts over a period of 20 years. This prompted us to review the literature on this subject. Among other aspects it was striking that, as in our case, most perforations described were in an area of localized hemorrhagic infarction in the apparently normal transverse or left colon (1,2,5,6,7,9,11). The explanations given for this phenomenon are prolonged direct pressure on the intussuscipiens by the swollen intussusceptum (6,7,9,10), sequential involvement of the blood supply of the distal bowel (7,10), excessive intraluminal pressure in young infants together with the usual enema pressure (1,6,10), kinking of the bowel wall or mesentery (6), colitis, anomalous blood supply and shock (1). To study the effect of strangulated intussusception we adopted and modified an animal model earlier described by Ravitch (14). In these experiments after laparotomy of a dog intussusception was produced instrumentally and strangulation was achieved by selective ligation of venes. We examined the specimens of 6 ileocolic and 4 ileoileal intussusceptions in 6 dogs with special interest in ischemic changes of the intussuscipiens.

Case report

A 17-month-old female child was transferred from another hospital for uncomprehended vomiting, abdominal pain and diarrhoea which existed 11 days. At surgical consultation she was pale and listless but afebrile. Examination of the abdomen revealed distension, decreased bowel sounds and hyperresonance. A tender sausage-shaped mass was felt in the left abdomen. Rectal examination was negative. Upright radiograph showed several gas-fluid levels with slightly dilated small bowel loops but no free gas. White blood count was 13.6 x 10.9/ml with a strong shift to the left. At barium enema the apex of an intussusception was encountered in the sigmoid colon. The intussusception was reduced to the left transverse colon where it stopped. The procedure was discontinued at this point and the operating room was prepared. No extravasation of barium was noticed. The contrast canister had not been elevated more than 100 cm above the tabletop. However shortly after this the childs condition deteriorated. At laparotomy 500 ml of contrast suspension was found in the peritoneal cavity. The apex of an ileocoecocolic intussusception protruded through a 10 cm long perforation in the

apparently normal splenic flexure. After manual reduction a small second perforation was found in the coecum. There was no evidence of a leading point. The perforations were oversewn, the peritoneal cavity was cleansed and an ileostomy was performed. The post-operative period was uneventful apart from a wound infection after closure of the ileostomy on the 24th postoperative day.

Materials and methods

The animals used were 6 beagle dogs weighing 12 to 15 kg. Under nembutal anaesthesia a midline laparotomy was performed. The vermiform appendix was freed from the last 5 cm of ileum. At 2 cm proximal to the ileocoecal valve 4 traction sutures were inserted. The ileum was seized by smooth forceps and inverted into the distal segment without causing damage to the intussuscipiens by any manipulation. A 15 cm long ileocolic intussusception was secured by 4 or 5 seromuscular stitches at the neck. All veins of the intussuscepted ileum were seperately ligated including those of the arcades at the ileocoecal junction and at the neck of the intussusception. All arteries remained intact. In three dogs an ileoileal intussusception was made in the same way. The wound was closed in one layer. After 6 hours, the dog still anaesthetized with normal blood pressure, the abdomen was opened again and the intussusceptions were taken out for histological examination. Multiple cross sections from the neck to the apex inclusive were made.

Results

The macroscopic aspect of all intusscipientes was that of normal colon or ileum. The mucosa of the entering layer was swollen, edematous and slightly hemorrhagic but its structure was grossly preserved. Although there was some edema and extravasation in the submucosa, it was of normal thickness. The inner circular muscle coat of the entering layer was greatly thickened. The outer muscle coat and the serosa were well preserved. The returning layer was found severely damaged. The mucosa of this layer showed variations in thickness and degree of damage from dog to dog. It was flattened with sloughed villi and with only a few crypts recognisible in an amorphous mass or it was edematous and hemorrhagic with still remnants of villi. The submucosa and the muscular layers showed edema and hemorrhage. The serosa showed extravasation of blood. All layers of the intussuscipiens were without changes with the exception of a slight round cell infiltration of the serosa. In 4 of 6

ileocolic and in 2 of 4 ileoileal intussusception however we found changes in the mucosa of the intussuscipiens. These changes were found on locations where there was a close contact between the intussusceptum and the intussuscipiens. On these locations we found edema, hemorrhage and sloughing of epithelium. We did not find any indication of impairment of the circulation of the intussuscipiens (Figure 1 and 2). A mechanincal cause seemed most likely.

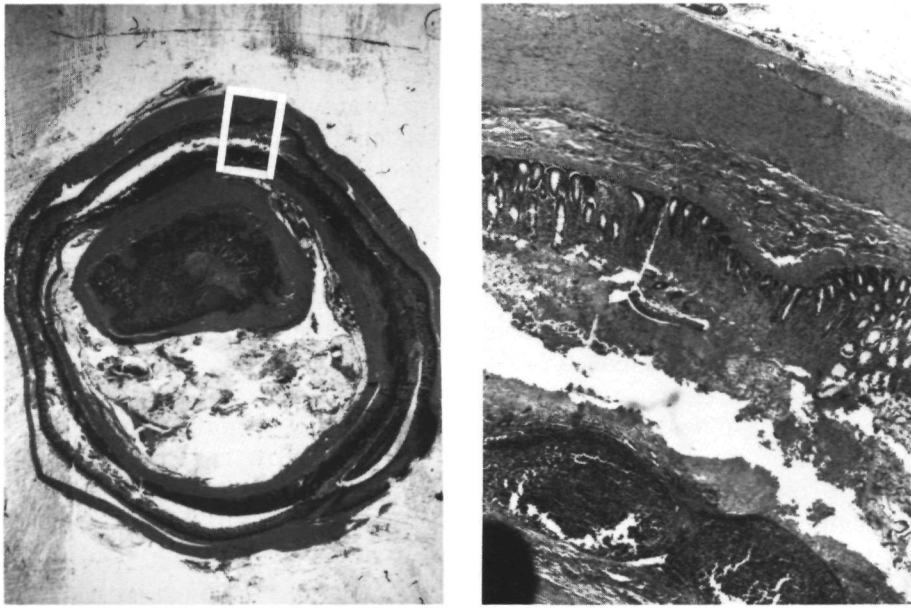

Fig. 1 and 2 (detail). Ischemic changes in the mucosa of the intussuscipiens in close contact with the intussusceptum.

Discussion

Patients with pallor, dehydration, lethargy and intermittent skin mottling seem to be at increased risk for perforation of the bowel during attempt at hydrostatic reduction of intussusception.

Especially infants up to 9 months of age with a long history (more than 36 hours) who have bloody stools and small bowel obstruction are at risk (7,9). However our case illustrates that older children can also have their bowel perforated. In many cases perforation occurs simultaneously with making the diagnosis at barium enema before any attempt at reduction can be made (6,7,9,12). On the other hand cases with perforation without extravasation during

barium enema with attempt at hydrostatic reduction, as in our patient, have been described (3,6). The apex of the intussusceptum can, at least temporary, block the perforation opening (15,11). Most perforations occur in that part of the colon surrounding the intussusceptum. A few cases have been reported in which the perforation was distal to the point were the apex of the intussusception was firstly seen at laparotomy or during barium enema (1,2). The apex of intussusceptions with perforation of the intussuscipiens invariable lies in the transverse colon or more distal. In our opinion in these intussusceptions the caecum often gets involved in the intussusceptum. In these cases there is a higher risk of vascular deprivation, by obstruction of the arcades of the hepatic flexure or of the superior mesenteric artery itself. On the other hand, after right hemicolectomy the vitality of the anastomosis also depends on the distal arcades, and usualy without any problem. As in our case a second perforation is often found, in some cases near the most distal one but more frequently near the neck of the intussusception in the caecum or ileum (1,2,5,6,7). It is striking how often the viability of the wall of the colon surrounding the perforation is mentioned as normal. We believe that in such cases and if the intussusception can be reduced manually, oversewing of the perforation in combination with an ileostomy is justified. Good results have been described earlier (1,5,6,8). These perforations do not make a resection necessary.

We found some indications in our animal model that local ischemic changes in the mucosa of the intussuscipiens can occur at places where a close contact with concomitant pressure between the intussusceptum and intussuscipiens exists. From other experimental studies in specimens of dogs and humans we know that the (sub)mucosa of the bowel is the most resistent layer to intraluminal pressure (4,13). Other explanations are less likely. After occlusion of more distal branches of the superior mesenteric artery, alone or in combination with an anomalous blood supply one would expect ischemic changes of all layers over a more extended area of the colon. Colitis, seldom reported, may have predisposed to perforation and so may shock and spastic pressure but we do not hold these factors completely responsible for these changes. In our opinion our findings give an indication that perforation of the intussuscipiens during attempt at hydrostatic reduction occurs through areas of local ischemic infarction on the basis of direct pressure by the intussusceptum.

Acknowledgement

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III.8

SONOGRAPHIC FEATURES OF INTUSSUSCEPTION

An experimental study

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Summary

In an animal model, the sonographic features of intussusception were studied. These sonographic features were correlated with the anatomical characteristics and histopathologic observations of strangulated intussusception. The complete sonographic picture of intussusception is pathognomonic. Its components are a central, round sonolucency, containing a moderate echodense area and surrounded by a half-moon shaped echodensity. This, in turn is surrounded by a sonolucent peripheral band containing narrow, concentric, echodense rings. From neck to apex these structures show characteristic changes in shape and dimension. Knowledge of these specific sonographic features is important in the light of future developments in the field of diagnosis and treatment of intussusception.
Keywords: Children, digestive system, gastrointestinal tract, ultrasound studies, intussusception.

Introduction

The clinical application of sonography in the diagnosis of gastrointestinal tract disorders has greatly expanded over the past decade. In several reports the diagnostic value of sonography in cases of intussusception in adults as well as in children has been emphasized (1-11). Ultrasound can be extremely useful, especially in atypical cases if an abdominal mass is found. The successful use of sonography as a means to control hydrostatic reduction attempts has recently been described (12,13).

The anatomical features of an intussusception are quite characteristic (Fig.1). The recipient bowel or intussuscipiens forms the outermost of three concentric cylinders. The invaginated bowel or intussusceptum consists of two cylinders. The innermost cylinder is called the entering layer while the middle cylinder is called the returning layer. The mesentery is invaginated between the entering and the returning layer. Sonography can show these characteristics and so differentiate intussusception from other diseases of the gastrointestinal tract such as primary and secondary cancer, lymphoma, Crohn's disease, inflammatory bowel disease, bowel infarction, radiation ileitis or hematoma of the bowel wall.

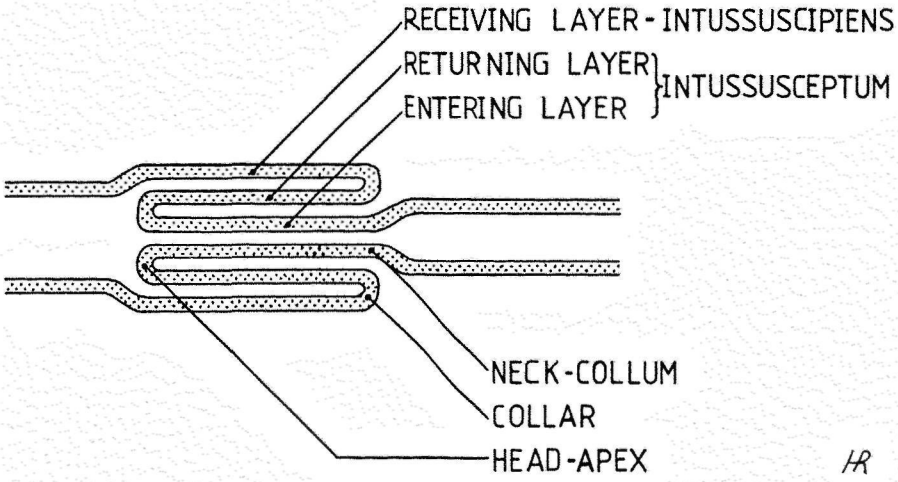

Fig. 1. Anatomical features of intussusception.

In most intussusceptions there is a simultaneous interference with the patency of the gastrointestinal tract and with the vascular supply of the intussusceptum. Most intussusceptions become increasingly strangulated as time passes. This results in venous compression, swelling of tissue and discharge of mucus with blood. Accordingly the sonographic features will also change with time. Knowledge of these changes is required to make the diagnosis with certainty.

We are unaware of reports of sonographic features of experimentally induced in vivo intussusceptions. In addition the relations between anatomic characteristics and sonographic features given in many reports seemed to us incomplete. Therefore, in an animal model, we observed the sonographic findings of intussusceptions in time. These findings were correlated with the anatomical characteristics and with pathologic observations. We undertook this study to determine the typical sonographic features produced by intussusception.

Method and materials

The animals used were eight beagle dogs weighing 12 to 15 kg. Under pentobarbital anaesthesia a midline laparotomy was per-

formed. The vermiform appendix was freed from the last 5 cm of ileum. At 2 cm proximal to the ileocecal valve four traction sutures were placed. The ileum was seized by smooth forceps and inverted into the distal segment. A 15 cm long ileocolic intussusception was secured by 4 seromuscular stitches at the neck. All veins of the intussuscepted ileum were separately ligated including those of the arcades at the ileocecal junction and at the neck of the intussusception. All arteries remained intact. In six dogs an ileoileal intussusception was made in the same way. All intussusceptions were fixed to the abdominal wall by transfixing sutures, at the same time marking the exact localization. After filling the abdominal cavity with isotonic salt solution to expel as much air as possible, the wound was closed in one layer. After 6 hours, the dog still anaesthetized with normal blood pressure, the abdomen was opened again and the intussusceptions were taken out for histological examination. Multiple cross sections from the neck to the apex inclusive were made. Histologic examination was carried out with special interest for the relative proportions of the component parts and signs of edema, haemorrhage, infarction and inflammatory reactions.

Serial sonographic examination was performed on each dog and on each intussusception immediately after and 2, 4 and 6 hours after surgery. Sonography was performed with a Toshiba Sonolayer Sal 77 B and a Philips SDR 1200. Standard transverse sections from neck to apex and longitudinal sections were taken. All sonograms were studied for features correlating with the anatomical characteristics of an intussusception. Sonograms were compared mutually for signs of edema, distinctness of layers and intraluminal mucus and fluid production.

Histologic layer thickness was measured at a magnification of X4 using an eyepiece micrometer accurate to 0.01 mm. Measurements of ultrasonographic images were made directly from the multiformat film using a dissecting microscope (magnification X4) and vernier calipers accurate to 0.03 mm. Layer measurements were taken from correponding areas of the ultrasound image and histologic section. Sonograms were also compared to "typical" pictures described in the literature.

Results

Transverse ultrasound scans (Fig. 2 and 3).

Transverse scans according to the longitudinal axis of the intussusception showed a circular mass. The mean diameter of both ileocolic

and ileoileal intussusception was quite constant in all animals from the neck to the distal portion inclusive. However, towards the apex the intussusception tapered down. The intussusception mass consisted of a sonolucent band with a constant width round on all sides. At the apex the width of this sonolucency rapidly increases, often asymmetrically. The outer boundary of this band was more irregular and less sharp than its inner boundary. One or two sharply delineated, narrow, concentric echogenic rings could be seen within this peripheral sonolucency over a large proportion of its circumference. These rings are best visualized ventrally and dorsally. Medially and laterally they are much less sharp or absent which leads to the conclusion that they mainly represent interfaces between layers. These rings measure 1 mm or less in width. Near the neck and in the proximal intussusception most of the times 1 ring was seen while the more distal portions showed three or four narrow rings.

The centre of the mass consists of two parts, an eccentric circular shaped sonolucency and a half-moon shaped echogenicity. The sonolucency was located ventral or ventrolateral in the area of the proximal and midportion of the intussusception. Nearer to the apex its localization varied and its diameter gradually increases. In the middle of the central sonolucency a small echogenic area is seen. At the neck of the intussusception the central echolucency can be seen entering the circular mass together with a echogenic zone making contact with the half-moon shaped echogenicity. At the very tip of the apex a collection of echoes of moderate density is seen in an area of sonolucency.

The half-moon shaped echogenicity surrounded the central sonolucency completely in the proximal intussusception in about half the cases. The size of the half-moon diminishes in the direction of the apex. Most of the times it was localized dorsal or mediodorsal in the central area of the circular mass.

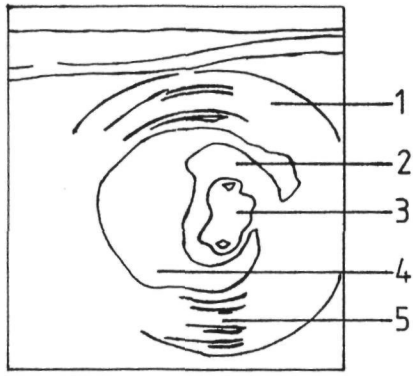

Fig. 2 and 3. Transverse ultrasound scan and diagram. 1 = Peripheral sonolucency, 2 = entering layer, 3 = lumen entering layer, 4 = mesentery, 5 = peripheral narrow concentric rings.

Longitudinal ultrasound scans (Fig. 4 and 5).

Scans along the longitudinal axis of the intussusception showed a mass with a tubular structure. Its maximal width was in accordance with the diameter of the circular mass found at transverse scanning. The image varied greatly. The most simple image was that of a sonolucent tube parallel to the abdominal wall with 1 to 4 parallel narrow echogenic stripes of at most 1 mm width. The image may also consist of two parallel sonolucent zones separated by a central echogenic area of varying width. Within the two sonolucent zones 1 to 4 parallel, narrow echogenic stripes could be seen.

Towards the apex the width of the two sonolucent zones increases. The area between the two sonolucent zones may also exist of an echogenic area in combination with a parallel sonolucent area localized ventrally, dorsally or in the middle of this echogenic area. This sonolucent area showed central echoes. The central echogenic area diminishes gradually from neck to apex. The central sonolucent area increases towards the apex. At the neck of the intussusception the central sonolucent area may be seen entering the tubular mass. At the apex a sonolucency is formed by a combination of an increased central sonolucent area and the two peripheral sonolucent zones.

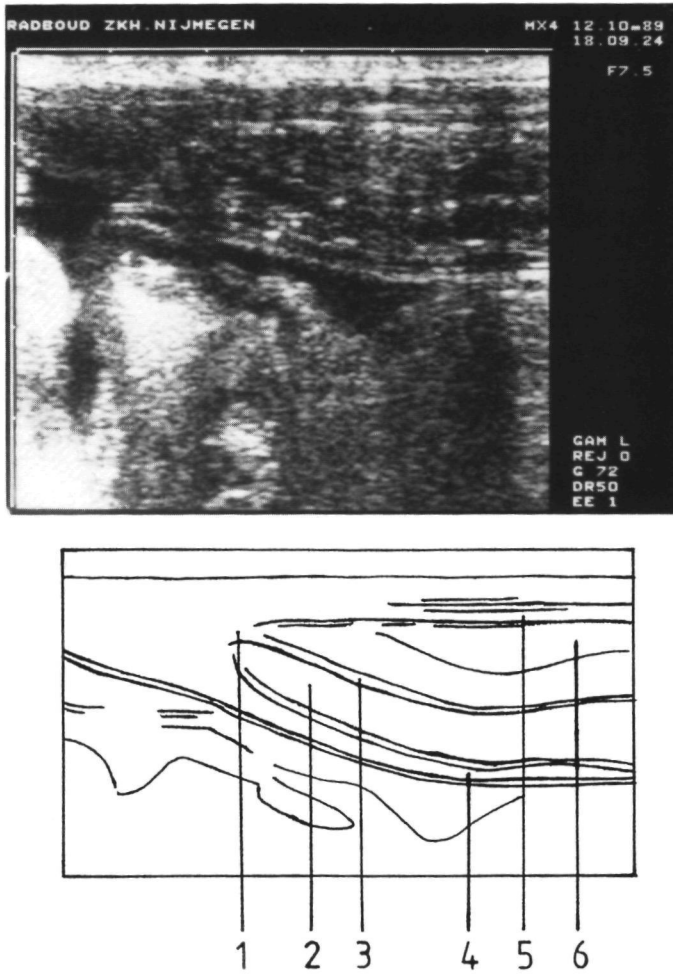

Fig. 4 and 5. Longitudinal ultrasound scan and diagram. 1 = Apex, 2 = entering layer, 3 = lumen entering layer, 4 = parallel narrow stripes, 5 = idem, 6 = mesentery.

Comparison of ultrasonographic images with histologic sections.

The receiving layer is stretched and less wide compared to the normal bowel wall but showed no generalized histologic changes. However the returning layer showed extensive changes. The mucosa of this layer showed variations in thickness and degree of damage. It was flattened with sloughed villi and with only a few crypts recognizable in an amorphous mass or it was edematous and haemorrhagic with remnants of villi. The submucosa and the muscular layers showed edema and haemorrhage. The subserosa showed extravasation of blood. The mucosa of the entering layer was swollen, edematous and slightly haemorrhagic but its structure was greatly preserved. Although there was some edema and extravasation in the submucosa, it was of normal thickness. The inner circular muscle coat of the entering layer was greatly thickened. The outer muscle coat and the serosa were well preserved.
The sonolucent peripheral band corresponds spatially to the combination of the wall of the receiving layer and the wall of the returning layer. The peripheral echogenic concentric rings correspond to the submucosa of the receiving and of the returning layer. The outermost sonolucent ring of the peripheral band corresponds to the muscularis propria of the receiving layer. The innermost sonolucent ring of the peripheral band corresponds to the muscularis propria of the returning layer. The intermediate sonolucent ring of the peripheral band is a combination of the mucosa of the receiving and the returning layer and sloughed mucosa and intestinal contents. Towards the apex the inner proportion of the peripheral sonolucent band increases in width.
The half-moon shaped echogenicity corresponds to the invaginated mesentery and its interfaces with the serosa of the returning and the entering layer. The interface with the entering layer can sometimes be seen as a narrow echogenic stripe, separated from the half-moon shaped echogenicity by a small layer of fluid. Towards the apex the half-moon shaped echogenicity gradually diminishes, corresponding to the decreasing size of the mesenteric mass.
The central sonolucency is similar to the entering layer and its central echoes are produced by its submucosa and the interfaces between mucosa and bowel contents. Towards the apex the edema and haemorrhage within the submucosa and within the muscularis propria in particular increases, resulting in a wider central sonolucency.

Ultrasonographic changes in time.

As time passes the central sonolucency increases in diameter (10-15%) because of extensive edema and haemorrhage in the submucosa and muscularis propria. Towards the apex the increase in diameter is more pronounced than near the neck of the intussusception, probably due to more room to distend. The innermost portion of the peripheral sonolucent zone also increases in time and more towards the apex. In the proximal part sometimes the two concentric narrow echogenic rings disappear or become one ring caused by stretching and distension plus compression.

Discussion

Over the years different ultrasonographic features of intussusception have been described. Weissberg et al. found a "target-like" appearance consisting of a sonolucent mass with dense central echoes to be suggestive of intussusception. Holt described a multiple concentric ring shadow, characteristic for intussusception. Morin reasoned that the "target-like" appearance was characteristic for transverse scans through the distal intussusception while the multiple concentric ring sign was characteristic for proximal scans. Montali described the longitudinal scan consisting of three parallel echolucent stripes delineating two echodense areas. Alessi specified the longitudinal appearance and called it the "hay-fork" sign. An "hourglass" appearance (6) and a "pseudo-kidney" sign (7) have been described as characteristic for intussusception. They are, as a matter of fact, the result of a combination of the exact transverse scan and the longitudinal scan of an intussusception (6). However, many reports were in some way inaccurate or incomplete. The development of better ultrasound instruments allowed the imaging of detailed anatomical characteristics of intussusception, not shown in the past. Recent experimental studies have been of great importance for the proper interpretation of ultrasonographic images (14).
The ultrasonographic pattern of intussusception is specific. Transverse scans show a sonolucent peripheral band with uniform thickness containing 1 to 4 narrow, concentric, echogenic rings. A (latero)ventral central circular sonolucency containing moderate dense echoes is surrounded by a (medio)dorsal, often half-moon shaped echogenicity, decreasing in size towards the apex. Longitudinal scans show tubular structures with alternating (0 to 3) echogenic and (1 to 4) sonolucent zones. Within the sonolucent zones several tubular echogenicities may be observed. From neck to apex

gradual differences in the size of the ultrasonographic structures may be seen.

We believe this sonographic pattern is pathognomonic for intussusception. Inflammatory and neoplastic bowel diseases show differences in the gross and detailed structure of their ultrasonographic pattern. Their echogenic centre usually is very small, longitudinal scans do not show a regular multizonal pattern, narrow rings and gradual longitudinal differences are usually absent and in neoplastic disease the outer sonolucency is more frequently eccentrically thickened.

Recent experimental work has made it possible to define the anatomical correlates of gastrointestinal ultrasound images (14). This interpretation takes into consideration the echoes produced by the tissue layers and the echoes produced by the interfaces between layers. Exact relating image and tissue is not possible due to tissue shrinkage and expansion during histologic processing, due to magnification of the ultrasound image and due to the fact that thickness on the ultrasound image is really a measurement of time. On experimental ultrasound images of the normal gastrointestinal tract three echogenic layers are distinguished. The superficial mucosa, the submucosa plus the acoustical interface between the submucosa and muscularis propria and the serosa and subserosal fat. Clinical transcutaneous techniques have been limited by the influences of intervening structures. Therefore we believe that in our experiments in the peripheral band on transverse scans only the thickest ultrasound images (submucosa/muscularis propria and serosa/subserosal fat) of the receiving and returning layer were seen.

In previous reports (10,11) ultrasonography was thought to provide data regarding potential hydrostatic reducibility of the intussusception. In the presence of layering or concentric rings or a thin outer sonolucent rim reduction was more likely while in the presence of tightly compressed central echoes and in the presence of a peripheral sonolucent band, thicker than 1.6 mm hydrostatic reduction was less likely. More clinical studies concerning the relation between the ultrasonographic appearance of intussusception and hydrostatic reducibility will be necessary. From our results one may conclude that the relative diameter of the entering layer, the thickness of the innermost proportion of the peripheral sonolucency and the number of narrow concentric echogenic rings are worth taking into account in these studies.

In our opinion the diagnosis of intussusception can be reached with certainty by ultrasonographic examination. Its successful use as a means to control hydrostatic reduction by normal saline solution

enema has been described. In the future the use of ultrasonography may be of great diagnostic and therapeutical importance.

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Intraperitoneal injection of Bordetella pertussis vaccine and Thiomersal failed to produce intussusception in mice

Keine Intussuszeption bei Mäusen nach intraperitonealer Injektion von Bordetella-pertussis-Vakzine und Thiomersal

Short communication

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Summary

No intussusception was caused by intraperitoneal injection of a combination of Thiomersal and Bordetella pertussis in 115 mice.
Keywords: Intussusception, invagination, mouse.

Introduction

We searched for an animal model of intussusception in infants and children. There is no species in which the incidence of intussusception is so high as to make it suited to study this condition. Hitherto in experiments the animal is laparotomized and the intestines are manipulated one way or another. Always the physiologic situation is violated severely.
In 1978 Blaskett et al. accidentally found a method to generate intussusception in mice by intraperitoneal injection of a combination of the preservative Thiomersal and a Bordetella pertussis suspension. They achieved intussusception in up to 30% of cases which would make this model a practical one (Blaskett 1978, 1979 and 1986). We wondered if this method could be as valuable in our hands following the indications of Blaskett as close as possible.

Material and methods

We used a random-bred Swiss (Cpb:Sc) mouse stock, SPF, kept under open conditions. Three to four weeks old mice were used with an weight range of 9 to 20 g. The animals, males and females not separated, were kept in groups of 10 in macrolon cages type III on sawdust and on a diet of RMH-pellets (Hope Farms, Woerden, The Netherlands) and tap water ad libitum. An adaptation time of 24 hours in a room with 20 to 22 °C temperature, atmospheric humidity of 50 to 60%, eight air changes per hour and artificial lighting during 12 hours daily was maintained. Bordetella pertussis, strain BP 21L3, provided by the RIVM 1), Bilthoven, The Netherlands , was grown on Bordet-Gengou plates and a suspension was made in casamino acids. Thiomersal (sodium ethyl mercurithiosalicylate) was supplied in a solution containing 0.01% in water.
For intraperitoneal injection, a mouse was anesthetized with ether and taped down on its back, after which its abdomen was swabbed with 70% alcohol. Anesthesia was maintained by means of an ether pad held over its nose. To avoid needle trauma to the intestines, a "tent" of midline abdominal skin was pitched up about a third of the way between the base of the sternum and the pubis. A 2-4 mm

patch of skin was removed, leaving the abdominal musculature intact. The injection needle, with loaded 2 ml syringe atttached, was inserted through the abdominal wall over the liver and 2 ml of the test material was discharged on the surface of the liver. The skin was then closed.
11 mice served as control group to check a correct technique, not traumatizing the intestines. In these mice 2 ml of a 0.9% NaCl solution was injected. All other 115 mice received 1.5 ml (150 µg) Thiomersal 0.01% simultaneously with different amounts of Bordetella pertussis bacteria in 0.5 ml. 52 mice were injected with 20 x 10.9 Bordetellae pertusses incubated for 30 minutes at 56 °C before injection to reduce the toxic side effects of Bordetella pertussis vaccine. 50 mice were injected with 10 x 10.9 Bordetellae incubated for 30 minutes at 36 °C and 13 mice recieved 2 x 10.9 organisms incubated for 30 minutes at 56 °C.
All animals that died were autopsied within 8 hours. 10 cm of ileum and the ileocaecal valve with 4 cm of colon were used for histopathological examination. Survivors were sacrificed for autopsy after 10 days.

Results

Neither in mice that died nor in survivors an intussusception was found. Histopathological examination showed no difference between mice that died and survivors. Congestion and oedema of all layers of small and large intestine, swollen mesenteric lymph nodes and sometimes small abscesses on the liver surface were found. Peyer's plaques were not strikingly hyperplastic compared to normal mice and to the control group.

Table 1. Outcome of intraperitoneal injection of Bordetella pertussis vaccine and Thiomersal preservative in mice.

Dose per mouse	Incubation for 30 minutes at	Number of deaths/total	Time of death
20 x 10.9 organisms	56 °C	8/52	1 x 3 days 7 x > 7 days
10 x 10.9 „	36 °C	50/50	50 x < 1 day
2 x 10.9 „	56 °C	–/13	–
control group	–	–/11	–

Discussion

Blaskett et al. (1978) attempted to increase the frequency of intussusception in mice by variing the dose of Bordetella pertussis as well as the dose of Thiomersal. They found a frequency of 7.3% in 177 animals injected with 100 to 190 µg Thiomersal and 1 to 60 x 10.9 Bordetellae pertusses per mouse. In a supplementary experiment with 17 mice they found an intussusception in 5 cases (Blaskett 1979 and 1986). Many factors can have caused the difference between their results and ours. Besides environmental factors the mouse stock or the Bordetella pertussis strain could have had different qualities. In fact it can not be excluded that in Blasketts experiments an unknown temporary intussusceptogenic factor was potentiated by Thiomersal as well as Bordetella pertussis suspension. Although the outcome of our experiment was negative we stress the importance in view of further research.

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CHAPTER IV

Summary and conclusions

In Chapter I the aim of the study before us is described.
We intend to draw attention to the uncommon but not rare problem of intussusception. We provide a review of literature data with emphasis on etiological, clinical, diagnostic and therapeutical aspects of intussusception. Our own observations and investigations aim to answer the following questions:

1. Which clinical features of intussusception bare diagnostic significance in intussusception? Which clinical features of intussusception are important from a diagnostic point of view for different age groups?

2. Can we provide guidelines on the choice of treatment of intussusception on the ground of clinical features? Must we provide different guidelines on the choice of treatment of intussusception for different age groups?

3. What can be the contribution of ultrasonography to the diagnosis of intussusception? What can be the contribution of ultrasonography to guidelines on the choice of treatment of intussusception?

4. Can we develop a practical, cheap and "physiologic" animal model to study intussusception?

The review of the literature in Chapter 2 makes it clear that in spite of more than a century of intense scientific interest in intussusception, – apart from the fact that evident progress in the field of treatment has been made – many questions still remain unanswered. In the course of this century, in western countries, the mortality from intussusception gradually dropped to almost nil. In addition to improved surgical and anaesthesiological techniques and better pre- and postoperative care, the fact that conservative treatment using standardized hydrostatic reduction has acquired the place of treatment of choice, is significant. Another important fact is that, due to better medical training, attainableness and public information the clinical picture is recognized much earlier in the

first and second echelon of health care. The use of non-invasive diagnostic tools with less side-effects, such as sonography, will be able to reduce the delay in treatment even further.
Many aspects of the treatment of intussusception are still subject to debate. It is still not clear how to differentiate those cases of intussusception which are best handled by primary operative treatment. Think of certain recurrent intussusceptions, intussusception on the basis of a malignancy, intussusception bound to be complicated by bowel perforation and intussusception that needs resection because of non-viability of the bowel. Another point of discussion is what is the most effective and at the same time safe technique for hydrostatic reduction. Manipulation of the intussusception tumour through the intact bowel wall, the elevation of the contrast canister, the number and duration of the attempts at reduction, the type of reduction medium (contrast, gas) and the administration of drugs during reduction are issues at discussion. The predisposition of infants, the male preponderance in childhood, the geographical variations and the predominant localization at the ileocecal junction are still unexplained phenomena. A clear proof of one of the many theories as to the pathogenesis of intussusception still has to be furnished. The incidence and the clinical relevance of transient physiologic intussusceptions is still unclear. Finally, a proper animal model to study intussusception, not violating the physiologic situation, is not at our disposal.

In Chapter III our observations in cases of intussusception and the results of our experimental work are described.
In the first part of Chapter III, the characteristics of our group of patients are described. In the district of Nijmegen many children with intussusception differed strikingly from the classical picture of intussusception given in textbooks and in publications concerning large series. In contradistinction to what has been stated in the past, ('Intussusception is relatively uncommon in malnourished children'! Hirschsprung), the weight of most children was under the fiftieth percentile. The percentage of small bowel intussusception was somewhat higher than reported in the literature. We found a low incidence of intussusception, especially in infants and young children. There were relatively far more children at school age. The delay in diagnosis was significantly longer and there was a very high percentage of leading points. These factors explain for the greater part the low hydrostatic reduction rate. Our conclusion is that it is important to be aware of geographical differences in these characteristics of intussusception evaluating the delay in diagnosis and the results of treatment.

In the second part of Chapter III, the results are described of a study of ten factors which, according to the literature, may play a role in an effort to provide guide-lines for the choice of treatment. A long duration of symptoms, vomiting, rectal bleeding, small bowel obstruction, ileo-ileocolic intussusception and the presence of a leading point were all significantly related to the failure of attempted hydrostatic reduction. However, according to logistic regression analysis, only "rectal bleeding" and "a duration of symptoms of more than 48 hours" contributed significantly to predict failure of hydrostatic reduction. We conclude that, besides the generally accepted contraindications, i.e. signs of peritonitis and of bowel perforation, the presence of rectal bleeding after a duration of symptoms of more than 48 hours is a contraindication for attempts at hydrostatic reduction.

In Chapter III.3 the appearance of neurological signs, early in the course of intussusception is separately discussed because this phenomenon is not adequately stressed in the literature. The presence of these signs was significantly correlated to a duration of symptoms less than 48 hours, rectal bleeding, bowel obstruction, intussusception with ileoileal component, dehydration, bicarbonate < 20 mmol/l and hydrostatic irreducibility. A definite conclusion concerning the origin of these signs, toxic metabolic, behavioral, reflexogen or sensory in nature, can not be drawn.

Chapter III.4 pays attention to a special group of children, to wit those with an intussusception that exists for more than 14 days. These intussusceptions are characterized by the (initial) absence of strangulation and thus of bowel obstruction and circulatory disturbances. Progress of intussusception may lead to acute symptoms. Most of these children are at school age. Diagnosis of intussusception is delayed, probably due to an unusual presentation. Compared with acute intussusception, symptoms consist of infrequent attacks of abdominal pain, sporadic vomiting and no, or small, changes in defecation. Marked weight loss and an abdominal mass assume diagnostic significance, in contradiction to bloody stools. Especially in the presence of an abdominal mass, ultrasonography can be of diagnostic value. An attempt at hydrostatic reduction is often unsuccessful. A high frequency of organic lesions precipitating intussusception warrants early surgical intervention.

In the fifth part of Chapter III intussusceptions in children 5 to 15 years of age are compared to intussusceptions in children under the age of 2 years. In the complete group of children with intussusception those at school age are numerically not unimportant. The

unusual presentation probably accounted for the fact that diagnosis is reached later. In over half the cases a predisposing factor is found, most of the times an organic lead point. Almost half the cases concern enteric intussusceptions. This warrants early surgical treatment. However, in the absence of contraindications, no child with intussusception with a large bowel component, should be disqualified from an attempt at hydrostatic reduction. After successful hydrostatic reduction scrupulous exclusion of an organic lesion, for instance by small bowel follow through examination, is required. If a chronic or chronically recurrent intussusception is suspected, surgical treatment is indicated since the incidence of organic lesions is high.

Chapter III.6 concerns diagnosis and treatment of intussusception in adults. Most of these intussusceptions originate in the small bowel. Due to the atypical clinical picture, usually, diagnosis of intussusception is not made before laparotomy. In addition to a high degree of suspicion, careful examination of plain abdominal radiograph and ultrasonography are of diagnostic importance. An organic lesion causing intussusception is nearly always found, which, in one third of cases, turns out to be a malignancy.
We conclude that adult intussusceptions with a large bowel component and in patients over 60 years old are best treated by primary resection. In adult intussusceptions originating in the small bowel an attempt at manual reduction followed by resection or enterotomy is justified. In patients with sigmoidorectal intussusception without endoscopically assessed evidence of malignancy, initial manual reduction is warranted thus avoiding a permanent colostomy.

In Chapter III.7 the problem of bowel perforation during hydrostatic reduction is discussed. Especially infants up to 9 months of age, with a duration of symptoms of more than 36 hours, with rectal blood loss and with small bowel obstruction are particularly at risk. Most perforations occur in an area of localized infarction in the normal transverse or left colon. We submitted the intussuscipiens of 10 intussusceptions, from an animal model, to histological examination in search of indications for the cause of this phenomenon. Ischemic changes in the mucosa were found on locations where there was a close contact between the intussusceptum and the intussuscipiens. At the same time no signs of impaired circulation were found in the remaining intussuscipiens. These findings indicate that perforation of the intussuscipiens during attempt at hydrostatic reduction occurs through areas of localized ischemic infarction on the basis of direct pressure by the intussusceptum.

Chapter III, part 8 deals with the sonographic features of intussusception. In an animal model exact sonographic pictures of strangulated intussusception were taken and described. The sonographic features were correlated with the anatomic characteristics and histopathologic observations of intussusception. The complete sonographic picture of intussusception is pathognomonic. Its components are a central, round sonolucency, containing a moderate echodense area (the entering layer). The round sonolucency is surrounded by a half-moon shaped echodensity (the invaginated mesentery). This, in turn is surrounded by a sonolucent peripheral band (the combination of the returning and the receiving layer) containing narrow, concentric, echodense rings. From neck to apex these structures show characteristic changes in shape and dimension. Knowledge of these specific sonographic features is important in the light of future developments in the field of diagnosis and treatment of intussusception.

In Chapter III.9 our endeavour to develop an experimental animal model to study intussusception is described. Besides economical and ethical deliberations the strive for a model with intact anatomy and physiology had priority. Although we found indications in the literature that it should be possible to cause intussusceptions in mice by intraperitoneal injection of Bordetella pertussis vaccine and Thiomersal, we failed to reproduce this. Further research, aiming at one of the many variables in this model, may reveal the cause of our failure.

In Chapter IV the preceding Chapters are summarized and the conclusions are presented.

In conclusion we want to emphasize that
1. it takes a high degree of clinical suspicion to reach early diagnosis in all cases of intussusception. The cardinal symptoms and signs of the past, i.e., colicky abdominal pain, vomiting, rectal blood loss and an abdominal or rectal mass are still important. However, one has to take into account that one or more of these cardinal symptoms and signs are often lacking. In several respects intussusception in our region differed from the classical picture. An important percentage of our patients are not the well-nourished children of the textbooks. Similarly an important proportion are older than 2 years of age. Frequently neurological signs such as altered consciousness, lethargy and generalized hypotonia are encountered early in the course of the disease. Chronic intussusceptions form a

substantial proportion of all cases of intussusception. Usually its clinical picture is not classical and characterized by marked weight loss.
In order to promote early diagnosis of intussusception we must emphasize that, in the absence of the classical symptoms and signs, neurological signs and weight loss are frequently found. A body weight under the fiftieth percentile and age over 2 years do not exclude intussusception. Once more we emphasize that early diagnosis is related to a benificial outcome of treatment. This applies to a greater extent to children of school age and for adults. The clinical picture in these patients differs strikingly from the classical picture of acute intussusception. It resembles that of chronic or chronically recurrent (partial) bowel obstruction. This clinical picture, in these age groups, should remind one of intussusception.

2. The treatment of choice of intussusception is conservative, to wit hydrostatic reduction. Absolute contraindications are signs of peritonitis and of bowel perforation. In the absence of contraindications no child with intussusception with a large bowel component should be disqualified from an attempt at hydrostatic reduction. In a number of cases regard must be paid to a higher than average probability of failure of hydrostatic reduction attempt. This is the case with patients with rectal bleeding and a duration of symptoms of more than 48 hours. The same accounts for patients with neurological signs and for patients with chronic intussusception (duration > 2 weeks). According to the literature, infants up to 9 months of age with a history of more than 36 hours who have bloody stools and small bowel obstruction should be treated extremely carefully because of the increased risk of bowel perforation during an attempt at hydrostatic reduction. In the above mentioned situations one has good reasons not to stubbornly persist at an attempt at hydrostatic reduction but to switch to surgical treatment in an early stage. Chronic and chronically recurrent intussusceptions are usually treated surgically, the more so as a high percentage of organic lesions can be found.
The treatment of choice is not the same for all age groups. The conditions necessitating extra caution in infants have been mentioned overhead. With age, the risk at intussusception due to an organic lesion increases. Besides, with age the risk that this etiologic organic lesion is a malignancy, increases. For this reason, after successful hydrostatic reduction in a child of school age, the evacuation radiograph must be examined carefully and an organic lesion must be ruled out, for instance by small bowel follow through meal. Adult intussusception must be treated surgically. Adult intussusception with a large bowel component and intussusception

in patients over the age of 60 are best to be resected primarily. In adult intussusception with its origin in the small bowel, manual reduction followed by resection or enterotomy is justified.

3. The complete transverse ultrasonographic picture of intussusception consists of a round central sonolucency with a moderate echodense center. This sonolucency is surrounded by a crescent shaped echodensity. This half moon is surrounded by a peripheral sonolucent band containing narrow, concentric, echodense rings.
Together with the longitudinal sonographic features this picture is pathognomonic for intussusception. The use of ultrasonography can contribute significantly to early diagnosis of intussusception, especially in atypical cases such as small bowel intussusception. Further investigations are needed to determine which sonographic features of intussusception may contribute to the choice of treatment.

4. A practical, cheap and "physiological" animal model for the study of intussusception is still lacking as our attempt to develop such a model failed. Our experiment as well as the data from the literature in this thesis may indicate the direction for future investigations.

Samenvatting en conclusies

In hoofdstuk I wordt het doel van de voorliggende studie aangegeven. De eerste doelstelling is aandacht te vragen voor het niet frequent voorkomende maar ook niet zeldzame probleem van de darminvaginatie. In een literatuuroverzicht worden daartoe de etiologische, klinische, diagnostische en therapeutische aspecten van invaginaties toegelicht. Naar aanleiding van klinische observaties en experimenteel onderzoek wordt getracht antwoord te geven op de volgende vragen:

1. Welke klinische kenmerken van invaginaties zijn uit diagnostisch oogpunt belangrijk? Welke klinische kenmerken van invaginaties zijn van diagnostisch belang in de verschillende leeftijdscategorieën?

2. Kunnen op grond van klinische kenmerken van invaginaties richtlijnen worden aangegeven voor de keuze van de therapie? Moeten voor de verschillende leeftijdscategorieën aangepaste richtlijnen voor de keuze van de therapie worden aangegeven?

3. Welke kan de bijdrage zijn van echografie voor de diagnostiek van invaginaties? Welke bijdrage kan echografie leveren aan de richtlijnen voor de keuze van de therapie?

4. Is het mogelijk een praktisch, goedkoop en "fysiologisch" diermodel te ontwikkelen voor de bestudering van invaginaties?

Het literatuuroverzicht in Hoofdstuk 2 maakt duidelijk dat ondanks meer dan een eeuw intensieve wetenschappelijke belangstelling voor invaginaties er naast evidente vooruitgang op het gebied van de behandeling nog vele vragen onbeantwoord zijn gebleven. In de loop van deze eeuw is in westerse landen de mortaliteit ten gevolge van invaginaties geleidelijk gedaald tot bijna 0%. Naast verbeterde chirurgische en anaesthesiologische technieken en betere pre- en postoperatieve zorg is het feit dat de conservatieve behandeling middels gestandaardiseerde hydrostatische reductie zich een plaats heeft verworven als therapie van eerste keuze, hiervoor verklarend. Daarnaast is belangrijk dat het ziektebeeld door betere bereikbaar-

heid van medici, door betere opleiding en door publieke voorlichting veel sneller wordt herkend in de eerste en tweede lijn van de gezondheidszorg. Het gebruik van niet invasieve minder belastende diagnostische middelen als echografie zal vertraging in behandeling in de toekomst nog verder kunnen terugdringen. Ten aanzien van de behandeling van invaginaties staan diverse zaken ter discussie. Hoe gevallen van invaginaties welke het meest gebaat zijn bij primaire operatieve behandeling, te onderscheiden is nog steeds niet duidelijk. Men denke hierbij aan sommige recidief-invaginaties, invaginaties op basis van een maligniteit, invaginaties waarbij gemakkelijk een perforatie ontstaat en invaginaties die om een resectie van avitale darm vragen. Ook de meest effectieve en tegelijkertijd veilige techniek van hydrostatische reductie is onderwerp van discussie. Hierbij staan manipulatie van de invaginatietumor door de intacte buikwand, hoogte van de reductie-druk, aantal en duur van de reductie-pogingen, aard van het reductie-medium (contrast, gas) en het gebruik van medicamenteuze ondersteuning nog ter discussie.
Tegelijkertijd bestaan aan het eind van de 20e eeuw ten aanzien van andere punten nog even grote vraagtekens als aan het eind van de 19e eeuw. De voorkeursleeftijd van 0 tot 2 jaar, de predispositie op kinderleeftijd van het mannelijk geslacht, de geografische verschillen en de overwegende localisatie ter plaatse van de ileocoecale overgang zijn nog altijd onverklaarde fenomenen. Een sluitend bewijs voor een van de vele theorieën met betrekking tot de pathogenese van invaginaties moet nog steeds worden geleverd. De incidentie en de klinische relevantie van kortdurende "fysiologische" invaginaties is niet duidelijk.
Een geschikt dierexperimenteel model waarin, onder zo fysiologisch mogelijke omstandigheden, invaginaties kunnen worden bestudeerd, staat ons niet ter beschikking.

In hoofdstuk III wordt verslag gedaan van eigen bevindingen bij patiënten met een invaginatie en van de resultaten van experimenteel werk.
In het eerste deel van hoofdstuk III worden de karakteristieken beschreven van het eigen patiëntenmateriaal. De kinderen met een invaginatie in de regio Nijmegen verschilden opvallend van het klassieke beeld van invaginaties zoals dat wordt beschreven in naslagwerken en publicaties over grote series uit bekende klinieken in de wereld. In tegenstelling tot wat vaak wordt verondersteld, had slechts een minderheid van de kinderen een meer dan gemiddeld lichaamsgewicht. Het aantal dunne darminvaginaties bleek wat hoger dan in de literatuur aangegeven. De incidentie van invaginaties in de regio Nijmegen is laag, met name onder kinderen jonger

dan 2 jaar. Kinderen in de schoolleeftijd vormden een relatief grote groep. De vertraging in het stellen van de diagnose was waarschijnlijk daardoor opvallend groot. Het percentage oorzakelijke organische afwijkingen was hoog. Deze factoren verklaren grotendeels ons relatief lage hydrostatische reductie percentage. Onze conclusie is dat het belangrijk is zich bewust te zijn van het bestaan van geografische verschillen in deze karakteristieke factoren alvorens een oordeel uit te spreken over de snelheid van diagnostiek en het resultaat van de behandeling.

In het tweede deel van hoofdstuk III worden de resultaten beschreven van de studie van 10 factoren die volgens de literatuur een rol kunnen spelen bij het opstellen van vuistregels voor de therapiekeuze bij invaginaties. Een lange klachtenduur, braken, rectaal bloedverlies, dunne darm obstructie, het ileoileocolisch invaginatietype en de aanwezigheid van een oorzakelijke organische afwijking bleken allen significant gerelateerd aan het falen van een poging tot hydrostatische reductie. Maar, volgens de logistische regressie analyse, droegen slechts "rectaal bloedverlies" en een "klachtenduur van meer dan 48 uur" significant bij tot het voorspellen van het falen van hydrostatische reductie.
De conclusie luidt dat naar onze mening, naast de algemeen aanvaarde contraindicaties, te weten tekenen van peritonitis en van een darmperforatie, de aanwezigheid van rectaal bloedverlies meer dan 48 uur na het begin van de klachten een relatieve contraindicatie vormt voor een poging tot hydrostatische reductie. Een dergelijke poging is dan weinig zinvol.

In hoofstuk III.3 wordt het optreden van neurologische verschijnselen, vroeg in de loop van een invaginatie afzonderlijk besproken gezien het feit dat deze verschijnselen in de literatuur te weinig onder de aandacht zijn gebracht. De aanwezigheid van deze verschijnselen bleek significant gecorreleerd aan een klachtenduur van minder dan 48 uur, rectaal bloedverlies, ileus, invaginaties met een ileoileale component, dehydratie, een bicarbonaat concentratie gelijk aan of lager dan 20 mmol/l en het falen van een poging tot hydrostatische reductie. Een conclusie ten aanzien van de oorsprong van deze verschijnselen – toxisch metabool, gedragsmatig, reflexogeen of sensibel – liet deze studie niet toe. Het diagnostisch belang van deze neurologische verschijnselen, die zo vroeg in de loop van de aandoening kunnen optreden, wordt benadrukt.

Hoofstuk III.4 vraagt aandacht voor een aparte groep kinderen, namelijk die met een chronische invaginatie. Deze invaginaties worden gekenmerkt door de (aanvankelijke) afwezigheid van stran-

gulatie en dus van darmobstructie en darmcirculatiestoornis. Progressie van de invaginatie kan een acute situatie tot gevolg hebben. Het betreft vooral kinderen in de schoolleeftijd. Waarschijnlijk ten gevolge van het atypische klinische beeld wordt de diagnose laat gesteld. Vergeleken met acute invaginaties bestaan de kachten uit relatief zeldzame aanvallen van buikpijn, sporadisch braken en geen of geringe veranderingen in de defecatie.
In tegenstelling tot rectaal bloedverlies, dragen het aanzienlijke gewichtsverlies van deze kinderen en de aanwezigheid van een weerstand in de buik bij tot het snel stellen van de diagnose. Met name bij aanwezigheid van een weerstand kan echografie van diagnostische waarde zijn. Een poging tot hydrostatische reductie is vaak zonder succes. Mede omdat vaak een oorzakelijke organische afwijking aanwezig is, is vroege chirurgische behandeling gerechtvaardigd.

In het vijfde deel van hoofdstuk III worden invaginaties bij kinderen in de leeftijd van 5 tot 15 jaar vergeleken met invaginaties bij zuigelingen en peuters. In de groep van alle kinderen met een invaginatie zijn de kinderen in de schoolleeftijd getalsmatig niet onbelangrijk. De afwijkende presentatie is waarschijnlijk de oorzaak van het feit dat de diagnose gemiddeld later wordt gesteld. In meer dan de helft der gevallen bestond een oorzakelijke factor, meestal een organische en in bijna de helft der gevallen ging het om een dunne darm invaginatie. Dit rechtvaardigt vroeg chirurgische ingrijpen maar in afwezigheid van contraindicaties behoeft men geen enkel kind met een invaginatie met een dikke darm component een poging tot hydrostatische reductie te onthouden. Na geslaagde hydrostatische reductie is het nauwkeurig uitsluiten van een organische afwijking, bijvoorbeeld door middel van een dunne darm passage onderzoek, vereist. Bij verdenking op een chronische of chronisch recidiverende invaginatie is het percentage organische afwijkingen bij deze kinderen zo hoog dat ook ondanks geslaagde hydrostatische reductie, chirurgische behandeling is geindiceerd.

Hoofdstuk III.6 gaat over de diagnostiek en behandeling van invaginaties bij volwassenen. Meestal vindt een dergelijke invaginatie zijn oorsprong in de dunne darm. Door het atypische klinische beeld wordt de diagnose vaak niet voor de laparotomie gesteld. Naast een hoge mate van klinisch verdacht zijn, zijn nauwkeurig onderzoek van de buikoverzichtsfoto en echografie van diagnostisch belang. Bijna altijd wordt een oorzakelijke organische afwijking gevonden die in een derde der gevallen een maligniteit blijkt te zijn. De conclusie luidt dat bij volwassenen invaginaties met een dikke darm component en invaginaties bij oudere patiënten (ouder dan 60

jaar) het beste kunnen worden behandeld door middel van primaire resectie. Bij invaginaties die in de dunne darm beginnen is een poging tot manuele reductie gerechtvaardigd waarna een resectie of enterotomie kan volgen. Ook bij sigmoidorectale invaginaties die niet lijken te berusten op een maligniteit, lijkt primaire manuele reductie verantwoord te zijn om aldus een definitief stoma te vermijden.

In hoofdstuk III.7 wordt het probleem van de darmperforaties tijdens hydrostatische reductie besproken. Vooral zuigelingen tot de leeftijd van 9 maanden, met een klachtenduur van meer dan 36 uur, met rectaal bloedverlies en met een dunne darm ileus lopen dit risico. De meeste van deze perforaties treden op in een klein, geinfarceerd gebied van het overigens normale colon transversum, descendens of sigmoideum. We onderwierpen het intussuscipiens van 10 invaginaties verkregen uit een dierexperiment aan een histologisch onderzoek op zoek naar aanwijzingen voor de oorzaak van dit fenomeen. Ischemische veranderingen in de mucosa werden daar gevonden waar een nauw contact tussen intussusceptum en intussuscipiens bestond terwijl de circulatie van de rest van het intussuscipiens ongestoord was. De conclusie is dat onze bevindingen een vingerwijzing opleveren dat perforatie van het intussuscipiens tijdens een poging tot hydrostatische reductie optreedt in gebieden van beperkte ischemische infarcering op basis van directe druk door het intussusceptum.

Hoofdstuk III, deel 8 handelt over het echografisch beeld van invaginaties. In een diermodel werden nauwkeurige echografische afbeeldingen van een gestranguleerde invaginatie verkregen en vervolgens beschreven. Daarbij werden de beelden gecorreleerd aan de anatomische karakteristieken en histopathologische kenmerken van een invaginatie. Het complete echografische beeld van een invaginatie bleek uniek. Het bestaat uit een centrale, ronde, echoarme zone met daarin een matig echorijk gebied (de "entering layer"). Het ronde echoarme gebied wordt omgeven door een echorijk gebied in de vorm van een halve maan (het geinvagineerde mesenterium). Hieromheen bevindt zich een perifere echoarme band (de combinatie van "returning" en "receiving layer") met daarin smalle, echorijke, concentrische ringen (submucosa en serosa). Van de hals naar de apex van de invaginatie vertonen deze beelden karakteristieke veranderingen in vorm en afmeting. Kennis van deze specifieke echografische kenmerken is belangrijk voor de vroegtijdige diagnostiek en de behandeling van invaginaties in de toekomst.

In hoofdstuk III.9 wordt onze poging beschreven om tot een diermodel te komen voor de bestudering van invaginaties. Naast economische en ethische overwegingen speelde daarbij het streven een rol de normale anatomie en fysiologie van het proefdier zoveel mogelijk intact te laten. Hoewel in de literatuur aanwijzingen werden gevonden dat bij muizen door middel van intraperitoneale injectie van een combinatie van Thiomersal en Bordetella pertussis vaccin invaginaties konden worden veroorzaakt, gelukte het ons niet dit te reproduceren. Verder onderzoek moet zich richten op een van de vele variabelen die mogelijk de oorzaak zijn van dit falen.

Concluderend kan men stellen dat
1. Voor het vroegtijdig stellen van de diagnose invaginatie een hoge mate van klinische verdenking noodzakelijk is. De van oudsher cardinale symptomen, aanvalsgewijze buikpijn, braken, bloedverlies per anum en een abdominale of rectale weerstand blijven belangrijk. Men dient er echter rekening mee te houden dat vaak één of meerdere van deze hoofdsymptomen ontbreken. In meer opzichten weken invaginaties in onze regio af van het klassieke beeld. Deze patiënten zijn meestal niet de goed doorvoede kinderen uit de boeken en in een belangrijk aantal der gevallen zijn ze niet jonger dan 2 jaar. Vroeg in het beloop van de aandoending treden bij een belangrijke groep patiënten neurologische verschijnselen als verlaagd bewustzijn, lethargie en gegeneraliseerde hypotonie, aan het licht. Chronische invaginaties vormen een niet onbelangrijk deel van alle invaginaties. In deze gevallen is het klinisch beeld meestal niet klassiek en wordt mede bepaald door aanzienlijk gewichtsverlies van het kind. We moeten dus benadrukken voor het bevorderen van het vroegtijdig stellen van de diagnose invaginatie, dat naast het ontbreken van een of meerdere der klassieke symptomen, het optreden van neurologische symptomen en gewichtsverlies frequent voorkomen. Kinderen met een lager dan gemiddeld lichaamsgewicht of een leeftijd boven 2 jaar vormen een zeer belangrijk deel van de patiënten met een invaginatie. Het zij benadrukt dat in een groot aantal der gevallen een hoge mate van klinische verdenking noodzakelijk is voor het vroegtijdig stellen van de diagnose invaginatie.
In versterkte mate geldt bovenstaande voor de kinderen op schoolleeftijd en voor volwassenen. Het klinisch beeld bij deze patiënten wijkt sterk af van het klassieke beeld van een acute invaginatie. In deze gevallen wordt vaak het klinisch beeld van een chronisch of chronisch recidiverende (partiële) darmobstructie gezien. Bij patiënten in deze leeftijdcategorieën dient men dan aan een invaginatie te denken.

2. De behandeling van keuze van invaginaties is conservatief, bestaande uit hydrostatische reductie. Absolute contraindicaties zijn tekenen van peritonitis en van darmperforatie. In de afwezigheid van contraindicaties behoeft men geen enkel kind met een invaginatie met een colon component een poging tot hydrostatische reductie te onthouden. Er zijn echter een aantal situaties waarin men rekening dient te houden met een meer dan gemiddelde kans dat de poging tot hydrostatische reductie mislukt. Dat is het geval indien rectaal bloedverlies wordt geconstateerd terwijl de klachten langer dan 48 uur bestaan. Verder indien neurologische verschijnselen bestaan. Bovendien is de kans op hydrostatische reductie klein indien de klachtenduur 2 weken of meer bedraagt. Op grond van literatuurgegevens dient men bij zuigelingen tot de leeftijd van 9 maanden met een klachtenduur van meer dan 36 uur, met rectaal bloedverlies en met een dunne darmileus, uiterst voorzichtig te werk te gaan wegens de kans op een darmperforatie tijdens de poging tot hydrostatische reductie. In al deze gevallen heeft men goede redenen om niet langdurig en hardnekkig vast te houden aan een poging tot conservatieve behandeling maar juist om vroegtijdig over te gaan tot chirurgische behandeling.
De behandeling van invaginaties is niet voor alle leeftijdscategorieën dezelfde. Onder welke omstandigheden men bij zuigelingen voorzichtig conservatief moet zijn, is hierboven aangegeven. Met het stijgen van de leeftijd neemt de kans toe dat aan de invaginatie een oorzakelijke organische factor ten grondslag ligt. Bovendien wordt met het stijgen van de leeftijd de kans groter dat deze oorzakelijke organische factor een maligniteit is. Daarom is het geindiceerd om na een geslaagde hydrostatische reductiepoging bij een kind op schoolleeftijd de ontledigingsfoto goed te bestuderen en een organische afwijking uit te sluiten, bijvoorbeeld door middel van een dunne darmpassage onderzoek. Chronische of chronisch recidiverende invaginaties leiden, mede door het hoge percentage organische afwijkingen, meestal tot chirurgische behandeling. Op volwassen leeftijd dient de behandeling van invaginaties chirurgisch te zijn. Invaginaties met een colon component bij volwassenen en invaginaties bij patiënten ouder dan 60 jaar kunnen het beste primair worden gereseceerd. Bij invaginaties met hun oorsprong in de dunne darm bij volwassenen is een poging tot manuele reductie gerechtvaardigd waarna een resectie of enterotomie kan volgen.

3. Het volledige echografische beeld van een invaginatie bestaat uit een centraal, rond echoarm gebied met een matig echorijk centrum. Dit echoarme gebied wordt omgeven door een halve-maan-vormige echorijke zone. Deze zone op zijn beurt wordt omgeven door een buitenste, echoarme rand waarin smalle concentrische echodense

ringen te zien zijn. Dit echografisch beeld is pathognomonisch voor een invaginatie. Toepassing van echografie kan een wezenlijke bijdrage leveren aan het vroegtijdig stellen van de diagnose invaginatie, met name in atypische gevallen zoals van dunne darminvaginaties.
Welke echografische kenmerken een bijdrage kunnen leveren aan de richtlijnen voor de keuze van therapie, behoeft nog nadere studie.

4. Het ontbreken van een praktisch, goedkoop en "fysiologisch" dierexperimenteel model voor de bestudering van invaginaties is nog steeds een probleem aangezien een poging tot het ontwikkelen van zo'n model mislukte. Dit experiment evenals de literatuurstudie over invaginaties bij dieren mogen de richting aangeven waarin verder moet worden gezocht.

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Curriculum vitae

11 November 1951	Born in Gemert
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Stellingen

behorend bij het proefschrift

INTUSSUSCEPTION

a clinical and experimental study

van

J.A.M. Reijnen

De referentiepunten bij de evaluatie van diagnostiek en behandeling van kinderen met een invaginatie dienen te worden gekozen in relatie tot geografische variaties in de karakteristieken van het patientenmateriaal.

Een poging tot hydrostatische reductie van een invaginatie bij een kind met meer dan 48 uur bestaande klachten en bloedverlies per anum is op statistische gronden af te raden.

Lethargie, een gedaald bewustzijn en hypotonie zijn frequent voorkomende, vroege tekenen van invaginaties bij kinderen.

Bij kinderen met wekenlang bestaande klachten van buikpijn en braken, met aanzienlijk gewichtsverlies en een abdominale weerstand dient men aan een chronische invaginatie te denken.

Invaginaties bij kinderen op de schoolleeftijd presenteren zich meestal op niet-klassieke wijze en moeten vaak chirurgisch worden behandeld wegens een hoog percentage organische afwijkingen en een hoog percentage dunne darminvaginaties.

Invaginaties bij volwassenen dienen chirurgisch te worden behandeld.

Perforaties tijdens hydrostatische reductie van invaginaties vinden meestal plaats in het intussuscipiens. Deze perforaties berusten waarschijnlijk op locale ischemische veranderingen op basis van druk van binnenuit door het intussusceptum.

Het is mogelijk met behulp van echografie de diagnose invaginatie met zekerheid te stellen.

Evenals bij het cutane maligne melanoom wordt de prognose van patienten met een maligne melanoom van de anorectale regio voornamelijk bepaald door het stadium op het moment van diagnose en nauwelijks door de behandeling.

(M.W.N. Ward, British Journal of Surgery, 1986; 73:68-69)

Milde stadia van ingegroeide teennagels (stadium 1 en 2 volgens Heifetz (1937)) dienen op niet-operatieve wijze te worden behandeld.

(British Journal of Surgery, 1989; 76:955-957)

De dagelijkse zorg voor de ongevalspatient in Nederland zou aanzienlijk in kwaliteit vooruitgaan indien hieraan hetzelfde budget zou worden besteed als aan de zorg voor slachtoffers van hypothetische rampen.

Het vermijden en behandelen van pijn is een van de belangrijkste condities voor de genezing van patienten met een posttraumatische dystrofie.

Ter bevordering van de kwaliteit van opleiding, onderwijs en onderzoek dient de opleiding van algemeen chirurgen geconcentreerd te worden in perifere opleidingsklinieken en de voortgezette opleiding, in deelgebieden van de algemene heelkunde, in universitaire klinieken.

Uit de incidentie en ernst van blessures, tijdens hockey opgelopen, kan niet worden geconcludeerd dat het hier een "gevaarlijke sport" betreft.

(Geneeskunde en Sport, 1981; 14: 13-17)

Het na de invoering van het kunstgras voortdurend gebruik van de term "hockeystick-vormige incisie" doet ten onrechte vermoeden dat deze incisies tegenwoordig kleiner zijn.

Het gebruik van bloedleegte duidt op een chirurgische bloedarmoede.

In tegenstelling tot de eerste stellingen staat deze laatste stelling tot het proefschrift als de voorraadkast van de huidige chirurgische verpleegafdeling op zondagmorgen: er is geen verband meer !

(Marank Ermers, 1990)

Nijmegen, 25 juni 1990